**Lung hernias** (alternative plural: herniae) are defined as a herniation of the lung beyond the confines of the thoracic cage. They are uncommon, mostly seen post trauma or thoracotomies.

**Clinical presentation**

Hernias which are symptomatic may cause dyspnea, chest wall pain or a visible or palpable chest bulge (most common in intercostal lung hernias). They may also be asymptomatic.

**Complications**

- incarceration: rare
- strangulation: rare
- spinal cord compression: single case report [9]

**Pathology**

Lung hernias are classified by their anatomic locations and the mechanism by which they arise (congenital or acquired).

**Etiology**

They can be either congenital or acquired in origin (classified by Morel-Lavallée in 1847):

- congenital
- acquired (most common)
    - spontaneous
    - pathological
    - inflammatory
    - traumatic
    - iatrogenic, e.g. post-thoracotomy incision [3]

**Location**

- cervical: ~35%
    - protrusion of the lung through the anterior region of the thoracic inlet where there is a space between the scalenus anterior and sternocleidomastoid muscles
    - mostly seen in elderly patients with emphysematous lungs with weak cervical fascia
- intercostal: ~70% (range 60-83%)
    - result of the weakening of the thoracic wall or abnormally elevated intrathoracic pressure (e.g. weightlifters, wind instrument players)
    - in post-traumatic cases, the lung herniation may occur immediately after the impact or years later
- diaphragmatic: extremely rare
- mediastinal: rare

**Treatment and prognosis**

Asymptomatic lung hernias may be managed by close observation. In symptomatic cases, immediate reduction and closure of the defect are indicated to prevent incarceration and strangulation [3].

Although lung hernias are rare and usually benign in nature, it is important for physicians to be aware that these entities do exist so that they are not alarmed when they are encountered. Knowledge of the benign nature of lung hernias will prevent the use of unnecessary invasive procedures and surgery.

Discussion

Chest pain is one of the most common complaints leading patients to the emergency department (ED). Chest pain has many causes, including gastrointestinal (GI) causes. Diaphragmatic hernia (DH) is one possible cause of chest pain, but more likely presents with abdominal pain (68%), bowel obstruction (39%) and pulmonary symptoms (37%).[1] DH are most commonly congenital in origin but less commonly are acquired after major trauma (blunt or penetrating chest or abdominal) or minor trauma (sneezing,

Visual case discussion

A 33-year-old woman with surgical history of gastric band and gastric bypass, 13 and 10 years earlier respectively, presented to the emergency department due to chest pain. It was a sudden, sharp, cramping, pleuritic pain at the base of the left hemithorax that started 3 h earlier, associated with nausea. Patient reported previous intermittent, spontaneous, self-limiting episodes of dull pain in the left hypochondrium since gastric bypass surgery. Her physical examination revealed worsening of

Questions and answers with a brief rationale true & false and / or multiple-choice questions

**Question 1**

*Question Type*: multiple choice

*Question Text*: About the etiology of diaphragmatic hernias… (Select the false).

*Answer Options*

- a)

Diaphragmatic hernias are most common as an acquired phenomenon.

- b)

The most common etiology of acquired diaphragmatic hernia is secondary to trauma which results in diaphragmatic rupture.

- c)

Iatrogenic causes following surgery are the second most common cause of acquired diaphragmatic hernias.

- d)

Iatrogenic causes following surgery are very rare, with mostly just case

Inverted intercostal hernias are uncommon, and even more so when comprised of soft tissue instead of lung parenchyma in the postoperative context. This report demonstrates a case in with such a

hernia was diagnosed through chest multidetector computerized tomography in a 48-year-old woman who presented to the emergency room with respiratory symptoms and tested positive for severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2). She had positive surgical history for left lower lobectomy with bronchoplastic procedure and mediastinal lymphadenectomy, due to an endobronchial typical carcinoid tumor a few years ago. Therefore, it is important for radiologists to be aware of the imaging characteristics of inverted intercostal hernias, to avoid diagnostic errors. Introduction

Inverted intercostal hernias are uncommon findings after thoracic surgical procedures, as the majority of postoperative hernias in the thoracic region are comprised of lung tissue. Soft tissue inverted intercostal hernias are better depicted through multidetector computerized tomography (MDCT) of the chest, and radiologists should be aware of its appearance to avoid interpreting it as another entity, such as a tumor, especially in cases of incarceration.

1. Download: Download high-res image (759KB)
2. Download: Download full-size image

Fig. 1. Unenhanced chest CT images (soft tissue window) in the sagittal (A) and axial (B) planes revealing inverted intercostal hernia characterized by protrusion of major rhomboid muscle and fat tissue of the left posterior chest wall (arrow) towards the pleural cavity through the fifth intercostal space. Coronal-oblique (C) maximum intensity projection CT image (bone window) depicting the widening of the fifth left intercostal space (asterisks). Axial-oblique (D) CT image (lung window) showing the previously described inverted intercostal hernia (arrow). No lung parenchymal opacities with typical features of COVID-19 pneumonia were encountered. In addition, there were imaging findings compatible with left lower lung lobectomy.

Case presentation

A 48-year-old woman visited our emergency department with a 10-day history of intermittent symptoms of dry cough and myalgia, which had worsened on the previous day. No fever or dyspnea was identified. The patient reported contact with family members who had been having similar symptoms. Her medical records included an open lung surgery performed five years earlier, which consisted of left lower lobectomy with bronchoplastic procedure and mediastinal lymphadenectomy due to an endobronchial typical carcinoid tumor. There were no relevant complaints related to the thoracotomy scar. On examination, the patient's vital signs were within normal ranges. Thoracic auscultation and complete blood count were normal; C-reactive protein level was only mildly elevated (1.07 mg/dL). Real-time reverse-transcription polymerase chain reaction of her nasopharyngeal swab tested positive for severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) nucleic acid, thereby confirming the diagnosis of coronavirus disease 2019 (COVID-19). Unenhanced chest MDCT showed no lung opacities with typical features of COVID-19 pneumonia. In addition to usual findings related to the left lower lung lobectomy, the CT scan incidentally detected a convex lens-shaped herniation with protrusion of soft tissue of the left posterior chest wall (including major rhomboid muscle and fat tissue of the deep chest wall) towards the pleural cavity through the posterior region of the fifth intercostal space, which was widened (Fig. 1). This condition is known as inverted intercostal hernia.

Discussion

Postoperative thoracic hernias, defined as protrusion of lung or soft tissue through a widened intercostal space, are rare, and most reported patients present with outwards intercostal hernia of the lung [1,2]. Inverted intercostal hernia of soft tissue of the chest wall is an unusual type, defined as inwards protrusion of soft tissue into the pleural space, which is very rarely reported in the medical literature [2,3]. It is believed that the muscle tension in the chest wall is usually sufficient to prevent a protrusion of its soft tissue toward the pleural cavity (which has negative pressure) via an intercostal space that has been widened in patients who have undergone chest surgery. Progressive muscle weakness and inadequate closure of the chest wall are possible factors that contribute to its occurrence [1], [2], [3].

As in the case reported by Torres et al [2], our patient was diagnosed with inverted intercostal hernia by chest MDCT, which revealed a protrusion of soft tissue of the chest wall (including muscle and fat tissue) toward the pleural space through a widened intercostal space (previous surgical access). MDCT scan with multiplanar and tridimensional reconstructions and maximum and minimum intensity projection images can be considered an important tool for diagnosis and characterization of intercostal hernias [4]. Ultrasonography and magnetic resonance imaging may be radiation-free alternatives to CT for evaluation of the chest wall [4,5].

Weissberg et al [1] list a few indications for surgical correction of intercostal hernias, such as increasing size, pain, signs of impending incarceration, such as difficulty to reduce the hernia, and cosmetic reasons. However, since the literature regarding the issue is scarce, there is no consensus regarding which patients should undergo operative treatment and which techniques should be implemented. Unlike our case, Torres et al [2] described a patient with chest pain at the inverted intercostal hernia site, who underwent a surgical reapproach consisting of exploratory thoracotomy via the widened intercostal space followed by careful closure of the thoracotomy incision with strong stitches to prevent hernia recurrence. Eventually, the incarceration of soft tissue into the widened intercostal space can mimic a chest wall tumor as reported by Iwata et al [3].

Conclusion

Inverted intercostal hernias are an uncommon finding in the postoperative setting. Medical practitioners, especially thoracic surgeons and radiologists, should be aware about this condition and recognize it on CT images.

Abstract

Peripherally inserted central catheter (PICC) lines are used routinely in neonates requiring prolonged drugs and parenteral nutrition and complications are usually infectious, mechanical, or thrombotic in nature. Spontaneous extravascular migration is extremely rare, and presentation with large volume chest tube output even rarer. We present a case involving a neonate on extracorporeal membrane oxygenation (ECMO) after congenital diaphragmatic hernia (CDH) repair and discuss management strategies.

1. Case presentation

A 38 week gestational age baby with prenatally diagnosed left sided congenital diaphragmatic hernia (CDH) with stomach, small bowel, colon, spleen and liver herniated underwent planned vaginal delivery. Endotracheal intubation was performed immediately post-delivery, however despite escalating ventilatory support, veno-arterial (VA) extracorporeal membrane oxygenation (ECMO) was required due to severe respiratory failure. Successful open repair of his

severe (CDH study group type C) left-sided CDH was performed on ECMO 48 hours later, with a left sided chest tube placed. Due to ongoing medication and total parenteral nutrition (TPN) requirements that could not be delivered in the circuit, on post-operative day (POD) 2, a 1.9 French single lumen peripherally inserted central catheter (PICC) line was placed in the left median cubital vein and position confirmed in the superior vena cava by chest X-ray (CXR) (Fig. 1).

1. Download: Download high-res image (338KB)
2. Download: Download full-size image



Fig. 1. Radiograph from POD 2. Placement of LUE PICC line in the area of the upper SVC.

The left chest tube was placed on suction intermittently over the first post operative week to encourage mediastinal shift, with minimal serous drainage. During the second post operative week, the amount of serous drainage from the left chest tube progressively increased and required continuous suction to maintain ECMO flows. Subsequently, a right sided chest tube was placed to evacuate a new effusion, with similarly large serous output. In addition, the efficacy of the vasoactive drugs seemed to be intermittent with escalating doses required over time. The combined (right and left) chest tube output reached a maximum of 1185 cc per day (>300 cc/kg/day) (Table 1) and appeared to be transudative in nature by laboratory analysis.

Table 1. Chest tube output from post-operative day 3–36.

| Post-Operative Day | Left Chest Tube Output (cc) | Right Chest Tube Output (cc) |
|---|---|---|
| 3 | 0 | 96 |
| 4 | 0 | 10.9 |
| 6 | 0 | 356 |
| 9 | 146 | 142 |
| 10 | 0 | 220 |
| 11 | 750 | 390 |
| 12 | 70 | 690 |
| 13 | 341 | 641 |
| 14 | 901 | 325 |
| 15 | 790 | 272 |
| 16 | 285 | 40 |

| Post-Operative Day | Left Chest Tube Output (cc) | Right Chest Tube Output (cc) |
|---|---|---|
| 17 | 1120 | 65 |
| 18 | 883 | 20 |
| 19 | 235 | 5 |
| 20 | 90 | 23 |
| 21 | 90 | 23 |
| 22 | 24 | 0 |
| 23 | 2 | 9 |
| 24 | 0 | 0 |
| 25 | 2 | 10 |
| 26 | 2 | 10 |
| 27 | 0 | 20 |
| 28 | 0 | 35 |
| 29 | 0 | 0 |
| 30 | 20 | 0 |
| 31 | 0 | 0 |
| 31 | 0 | 0 |
| 32 | 0 | 0 |
| 33 | 0 | 0 |
| 34 | 0 | 0 |
| 35 | 3 | 20 |
| 36 | 0 | 0 |

CXR on POD 22 showed the PICC in a different location, and a contrast study noted that the tip was in the left chest cavity (Fig. 2, Fig. 3, Fig. 4).

1. Download: Download high-res image (305KB)
2. Download: Download full-size image

Fig. 2. Radiograph from POD 22 at 00:23. LUE PICC line expected location at level of the superior cavoatrial junction.

1. Download: Download high-res image (337KB)
2. Download: Download full-size image

Fig. 3. Radiograph from POD 22 at 09:32. Contrast injection through LUE PICC line indicating extravascular extravasation.

1. Download: Download high-res image (319KB)
2. Download: Download full-size image

Fig. 4. Radiograph from POD 22 at 12:28. LLE PICC line located in mid-right atrium.

The PICC was removed and a new one placed in the left saphenous vein (Fig. 4).

Chest tube output rapidly declined to less than 50 cc total (15 cc/kg/day). He was weaned off ECMO support and decannulated on post operative day 37. He was subsequently extubated and successfully weaned off oxygen support.

## 2. Discussion

Although the use of PICC lines have made a tremendous impact in the field of neonatal intensive care, they have also been shown to present with associated life-threatening complications, including infections, extravascular infiltrations or leak and thromboses [[1], [2], [3], [4]]. A five-year analysis of a single-center Level III Neonatal Intensive Care Unit in British Columbia predicted the incidence of PICC line related pleural effusions to be 0.4 per 1000 days of catheterization [5]. For decades, it has been recognized that rare, but extremely severe complications of PICC lines can also include cardiac tamponade and pleural effusions, however progressive refinements in the catheters have resulted in more flexible materials and fewer incidents [6]. A 2005 case report presented a neonate with a hydrothorax resulting from a central line that migrated out of position and led to cardiac and respiratory failure [3]. Another case noted a perforation through the pulmonary artery and bronchus to eventually drain contents into the lung [7]. An additional case report calls attention to the need for frequent monitoring of line placement following a neonate that had TPN draining into the retroperitoneal cavity following erosion of the catheter [8]. In summary, there have been various reported cases in which catheters in neonates become misaligned or eroded and ultimately lead to confusion for the clinical team, as in our case where the issues began much later after insertion leading to diagnostic confusion.

The specific etiology of the PICC line's extravasation in this case remains a mystery. Potential causes have been hypothesized in the literature and include excessive movements of the upper extremities and/or flushing of the catheter too rigorously by the clinical team [7]. However, the former is unlikely in this case given the patient's post-surgical sedation and dependence on ECMO. Other possibilities

include erosion of the catheter, leading to incomplete delivery of the contents to the desired location, such as vasoactive drugs in our case. This erosion hypothesis seems more likely given the clinical presentation of gradual decline, rather than immediate failure. Although the cause of this complication is still unknown, it is of the utmost importance that careful consideration of PICC line migration is considered in future cases when newborns present with copious chest tube outputs of unexplained etiology.

Chest tube placement is a nearly ubiquitous practice in thoracic surgery to ensure proper cardiopulmonary function [9]. Management of a chest tube requires careful oversight from medical and nursing staff to ensure the area remains patent and clean [10]. Potential etiologies for high chest tube outputs include pleural effusion, chylothorax, hemothorax, or rare cerebral-arachnoid pleural fistula [11]. General workup for high chest tube output commonly includes inspecting and characterizing the effluent, which was performed in this case to find it was transudative in nature [10,11]. Further workup if no clear etiology can be identified should be with chest radiography, which was performed and noted in Fig. 1, Fig. 2, Fig. 3, Fig. 4. Chylothorax is the most common culprit in neonates with large volume pleural effusions and drainage, however that occurs in patients with enteral feeding unlike our case with TPN.

Patients suffering from CDH often undergo intervention with mechanical ventilation, glucocorticoid administration, extra-corporeal membrane oxygenation, central line placement, and surgical repair [2,12]. The main complication of interest in this discussion is related to the PICC line, however, this case also presented with a long run of ECMO, beginning early on day of life 1. Experience from our institution has shown that long runs of ECMO in even the most severe cases have had excellent outcomes with survival rates of more than 50% [13]. In this case, it is likely that the PICC complication resulted in an extra 10 days of ECMO.

In conclusion, this case report calls attention to the need for careful monitoring of catheter placement and structural integrity. Extravascular PICC line migrations in neonates have been reported to cause complications outside of the initial placement period, and in rare cases continue to be a problem up to multiple weeks post-catheterization. In the setting of unexplained large volume chest tube output in neonates, we propose that careful consideration be placed on the position of central access. Furthermore, simple chest radiography might not be sufficient to diagnose extravasation and interrogation of central lines (i.e. contrast studies) may be warranted to diagnose such complication.

Abstract

Traumatic thoracic or chest wall hernias are relatively uncommon but highly challenging injuries that can be seen after a variety of injury mechanisms. Despite their description throughout history there remains scant literature on this topic that is primarily limited to case reports or series. Until recently, there also has been no effort to create a reliable grading system that can assess severity, predict outcomes, and guide the choice of surgical repair. The purpose of this article is to review the reported literature on this topic and to analyze the history, common injury mechanisms, likely presentations, and optimal management strategies to guide clinicians who are faced with these challenging cases. We also report a modified and updated version of our previously developed grading system for traumatic chest wall hernias that can be utilized to guide surgical management techniques and approaches.

Historical perspective

Traumatic thoracic injuries resulting in chest wall defects and associated herniation of lung or other thoracic contents are a relatively uncommon but highly challenging injury. Although these are typically associated with high velocity vehicular trauma in the modern era, they have been identified and described throughout known history. Severe chest wall injuries can also result from widely disparate mechanisms including large animal attacks, knife or ballistic injuries, and fall from heights,

Epidemiology & etiology

Chest wall hernias are most frequently post-traumatic (approximately 80%), but there are many other known causes: iatrogenic (e.g. post-operative), spontaneous (e.g. after coughing), infectious, congenital, and neoplastic.[18] For all etiologies the frequency of reported chest wall hernias is exceptionally low, amounting to case reports and abbreviated case series only.[19] Even during episodes of intense conflict the reported prevalence has been low; the Army Surgeon General reported 7 instances

Biomechanics

Chest wall hernias are associated with a loss of chest wall musculoskeletal integrity, and commonly contain both bony and musculofascial defect. However, chest wall hernias can be due to disruption of only the chest wall musculature, even if the bony structures of the chest wall are intact. It is notable that chest wall hernias with isolated disruption of the bony chest wall without injury of the chest wall musculature have not been described. The muscular injury may be a simple laceration,

Presentation, physical exam findings, and diagnosis/classification

Most patients with chest wall hernias are asymptomatic, especially if the etiology of the hernia is nontraumatic. They rarely experience shortness of breath or dyspnea. Patients may first notice a small area of pain at the location of the hernia, and some patients can have a bulge that appears with forced inhalation/exhalation, Valsalva or excessive coughing. The bulge is often soft and may or may not be reducible, and typically is located at the site of local trauma or chest wall disruption.

Treatment options

Due to the rarity of the condition, there is currently no standard recommendations on the management of chest wall hernias, and they are typically managed on a case by case by the individual surgeon. Treatment of these injuries can be managed either medically or surgically depending on the etiology, anatomic position, and symptomatology. Historically, acquired, non-traumatic or congenital hernias were managed non-operatively. In addition to recommendations of avoiding triggers that would

Outcomes/complications

Many of the postoperative complications of chest wall hernia repairs are not unique complications to the surgery. As chest wall hernias are overall a rare entity and outcomes after surgical repair/reconstruction have not been widely reported, the true incidence of individual complications is not well known. Perioperative complications include wound infections, pneumonia, failure to wean off ventilator/reintubation, recurrent hernia, prolonged air leak, and even death.[19] Given the nature of the

Summary

Traumatic chest wall hernias are a rare but challenging entity that may be seen after a variety of injury mechanisms.

**TOPIC:** Disorders of the Mediastinum

**TYPE:** Medical Student/Resident Case Reports

**INTRODUCTION:** Hiatal hernias are broadly divided into sliding and para-esophageal hernias. Sliding hernia is characterized by displacement of the gastroesophageal junction above the diaphragm.Mechanical problems caused by the hernia include respiratory complications like atelectasis, micro-aspirations & respiratory failure resulting from compression of the lung. We present a case of respiratory failure caused by retrocardiac hiatal hernia & its successful management with ultrasonic chest-physiotherapy (CPT).

**CASE PRESENTATION:** Our patient is a 68years old lady, who presented with complaints of shortness of breath & drowsiness for 3 days. She had similar complaints 2 months ago when she was diagnosed with sliding hiatal hernia with aspiration pneumonia. On presentation, auscultation of chest revealed diminished air entry in bilateral bases, but it was worse on right side. Labs showed WBC 10000, BUN 20, creatinine 1.8, ABG: pH 7.31/pCO2 61/pO2 60, lactate 5.9 & blood culture grew gram positive cocci in pairs. CXR showed right sided basal infiltrate, raised diaphragm & atelectasis with mucus plugging changes. CT chest revealed the preexisting large retrocardiac hiatal hernia. Barium swallow test showed severe thoracic esophageal dysmotility which is a contraindication to hernia repair.She was oxygenated with high flow non rebreather mask & intermittent BiPAP. IV fluids and antibiotic coverage with vancomycin & cefepime were given. She received 4 times a day regiment of ultrasonic CPT, acapella flutter valve & nebulization with salbutamol and ipratropium. Bronchoscopy was deferred due to anticipated complications of the procedure.Gradually patient improved clinically & her oxygen requirement decreased. There was improvement on CXR. She was discharged with instructions for aspiration prevention & is scheduled to follow up for PEG gastropexy.

**DISCUSSION:** Our patient was diagnosed to have type 1&2 respiratory failure due to aspiration pneumonia, atelectasis with mucus plugging & raised diaphragm- all complications of hiatal hernia. Surgery & bronchoscopy are used to treat respiratory complications of hiatal hernia. However certain scenarios may arise where these procedures cannot be performed. There are various upcoming non-invasive techniques which help resolve the respiratory compromise in such situations. Among them is ultrasonic CPT, where a hand-held vibratory device is used to simulate hand percussion during CPT. We can also nebulize the patient with agents like acetylcysteine before CPT to help loosen secretions. These methods help remove mucus plugs & improve oxygenation.

**CONCLUSIONS:** Ultrasonic CPT with other breathing treatments can be used to manage respiratory complications of hiatal hernia. This helps avoid invasive procedures like bronchoscopy and their complications. Hence more studies are required to encourage their use in situations where invasive procedures are contraindicated.

Abstract

Acute chest pain resulting in spontaneous idiopathic hemomediastinum is a rare, potentially life-threatening occurrence. Acute chest pain is a common chief complaint of patients, accounting for 2.4%–6.0% of adult emergency room visits. The clinician's differential diagnoses for acute chest pain rarely include complications of hiatal hernias. An 83-year-old male presented with acute chest pain and was emergently diagnosed with hemomediastinum secondary to spontaneous gastric mesenteric vessel rupture due to a non-strangulated hiatal hernia after physical exertion.

## 1. Introduction

Idiopathic spontaneous hemomediastinum is infrequently encountered in the emergency department (ED). The three types of spontaneous hemomediastinum that have been described previously are as follows: first one is due to bleeding disorders such as hemophilia, anticoagulant use; secondary one is due to mediastinal tumors such as teratomas and thymomas, that may include other organs and blood vessels, and the third one (idiopathic) is due to sudden increase in intrathoracic pressure (*e.g.*, during coughing, sneezing, vomiting or sudden sustained hypertension)[1], [2], [3]. The most commonly encountered large vessel pathology resulting in hemomediastinum is due to aortic aneurysm dissection[1]. The case described herein is a rare case of idiopathic spontaneous hemomediastinum presenting as acute chest pain.

## 2. Case presentation

An 83-year-old Caucasian male presented to the ED via ambulance with a chief complaint of acute chest pain after performing sixty push-ups prior to arrival. He stated that he maintained an active life style and performed sixty push-ups in the morning and at night as part of his daily exercise regimen. The patient stated right-sided, sharp chest pain radiating to his back that occurred at rest and was not reproducible. The pain increased with deep inspiration and was associated with mild dyspnea and dizziness. The patient had a significant past medical history of hypertension, hiatal hernia, benign prostate hyperplasia and a significant past surgical history of prostate biopsy. The only reported prescribed medication was lisinopril and hydrochlorothiazide 20/12.5 mg tablet that was taken daily.

Upon ED arrival, the patient's vital signs were as follows: blood pressure of 87/60 mmHg, heart rate 67 beats per min, respiratory rate 18, afebrile, and $SpO_2$ 100% on two liters nasal cannula. On physical examination, he was pale however well nourished. The patient was noted to have decreased bilateral breath sounds. He did not exhibit distended neck veins, muffled heart and/or murmurs/gallops, tracheal deviation, stridor, blood pressure or radial pulse discrepancy in his upper extremities, nor a palpable pulsatile abdominal mass.

An additional peripheral line was placed, cardiac monitoring, pulse oximetry and a normal saline 1 L bolus were provided. An electrocardiogram (EKG), complete metabolic profile, troponin-T, pro-brain natriuretic peptide, complete blood count, liver function tests (AST/AGT), coagulation profile, type and screen were performed. Additionally, the patient was kept “nothing by mouth status” for the possibility of surgical intervention. Emergent radiographic imaging included portable chest X-ray and CT scan of the chest with contrast (aortic dissection protocol).

EKG and blood work were noncontributory. The portable chest X-ray (Figure 1) demonstrated a large hiatal hernia, a tortuous aorta and chronic changes. CT of the chest demonstrated a 4.5 cm dilated ascending thoracic aorta without evidence of dissection or distal thoracoabdominal aneurysm (4 cm descending aorta) (Figure 2). Interestingly, a large collection concerning hematoma was occupying most of the hiatal hernia (17 cm × 10 cm on the axial images × 15 cm coronal images) (Figure 3, Figure 4). Additionally, a small portion of stomach was found to protrude superiorly into the inferior aspect of the hiatal hernia with extravasation of amorphous hyperdense/hemorrhagic fluid, concerning gastric mesenteric vessel rupture. Immediate cardiothoracic and intensive care consults were obtained.

1. Download: Download full-size image

Figure 1. Portable chest X-ray demonstrating a large hiatal hernia and tortuous aorta.

1. Download: Download full-size image

Figure 2. Axial view CT with contrast demonstrating 4 cm descending thoracic aorta.

1. Download: Download full-size image

Figure 3. Axial view CT with contrast demonstrating large hiatal hernia with extravasation of contrast (circle).

1. Download: Download full-size image

Figure 4. Coronal view CT with contrast demonstrating large hiatal hernia with 15 cm hemomediastinum (A).

The patient was reassessed for clinical deterioration after return from CT scan. He was pain free, with repeat vitals: blood pressure of 137/72 mmHg, heart rate 66 beats per min, respiratory rate 23, afebrile and $SpO_2$ 100%. Cardiothoracic surgery recommended nasogastric tube insertion and decompression as well as close monitoring in the hospital. A small amount of gastric aspirate was obtained from the nasogastric tube. The patient was admitted and subsequently declined any further testing (esophagram) or surgical intervention. He left the hospital two days later without incident.

3. Discussion

Acute chest pain is a common ED chief complaint[4], [5]. Previous studies have reported that 54.5% of patients had neither clinical nor EKG evidence of an acute coronary syndrome, but 12.5% were categorized as having other life-threatening pathologies[4]. The differentiation of ischemic versus non-ischemic chest pain can be an especially daunting task when patients present with acute chest pain[6]. The differential diagnosis of acute chest pain is wide. Proper triage and physical examination including past medical/surgical history remain paramount. The patient's chief complaint in this case prompted our team to pursue an aortic (thoracic) dissection. The initial portable X-ray revealed a large hiatal hernia as well as a tortuous aorta. The CT chest provided the rare diagnosis of idiopathic spontaneous hemomediastinum secondary to a mesenteric vessel rupture in the patient who had a known hiatal hernia. Since the patient did not suffer recent trauma or falls, the vessel rupture most likely occurred during physical exertion. Push-ups increase intrathoracic pressure and may have contributed to this patient's diagnosis. Intestinal strangulation or viscous perforation was not observed in this case, although has been reported previously[6].

This case illustrates the importance of high suspicion of underlying pathology secondary to clinical gestalt, mechanism of injury, patient's medical history and need for early specialty consultation. It additionally demonstrates the vitality of appropriate emergent radiographic imaging, in this case CT, to reduce morbidity and mortality.

Introduction and imporatnce

Introduction: Bochdalek's hernia (BH) is a congenital diaphragmatic hernia predominantly diagnosed in the pediatric population but infrequently found in adults. This paper presents a unique case of an adult patient with a left-sided BH accompanied by gastric volvulus and an intrathoracic kidney.

Case of presentation

A 21-year-old male presented with abdominal pain and vomiting. An MDCT scan revealed a twisted stomach, spleen, and kidney herniated into the chest due to left diaphragmatic eventration. Surgery involved untwisting the stomach, relocating the organs, and removing the hernia sac.

Discussion

Bochdalek hernias (BHs) are rare conditions in which abdominal organs move into the chest due to defects in the diaphragm. BH usually occurs on the left side and can be triggered by factors such as pregnancy, obesity, or trauma. Symptoms can vary from abdominal pain to chest discomfort, and diagnosis can be challenging. Imaging tests such as CT scans are essential for accurate diagnosis. In adults, the BH can contain various organs, such as the spleen and kidney. Rarely, BH can be associated with an ectopic kidney located inside the chest cavity. In some cases of BH, there is a risk of complications such as gastric volvulus, where the stomach twists on itself, leading to potentially serious symptoms such as severe abdominal pain and vomiting.

Conclusion

This case underscores the severe risks of BH in adults, such as gastric twisting and blockage, necessitating urgent surgery. Timely diagnosis and surgical intervention are crucial for preventing life-threatening outcomes. More research is needed to improve the management of this rare condition.

## 1. Introduction

Bochdalek's hernia (BH) is a congenital diaphragmatic hernia that arises from the persistence of the pleuroperitoneal canal due to the incomplete fusion of the pleuroperitoneal folds. This anomaly is commonly found in the pediatric population but is infrequently diagnosed in adults [1]. The exact prevalence of BH is uncertain, but the incidence of all diaphragmatic hernias in adults ranges from 0.17 % to 6 %. There are fewer than 100 reported cases of BH in adults in the literature [2]. BH in adults typically manifests on the left side of the diaphragm due to the earlier closure of the right pleuroperitoneal canal and is often discovered incidentally [1]. However, its clinical presentation can vary and can include both abdominal and chest symptoms [1]. Although infrequent, complications of BH in adults, such as gastric volvulus and strangulation, can occur. These constitute life-threatening conditions that require immediate emergency surgery [3]. An intrathoracic kidney, considered one of the rarest displacements, has a prevalence of less than 1 in 10,000 cases. Despite the increasing recognition of this condition in the literature, its occurrence in BH is considered extremely rare based on the available database [4,5]. In this study, we present a successful surgical case involving an adult with a left-sided BH accompanied by gastric volvulus and an intrathoracic kidney.

This work has been reported in line with the SCARE criteria [6].

## 2. Case presentation

A 21-year-old male Bangladeshi patient presented with diffuse abdominal pain that had started three days prior and had become severe in the last 24 h. The pain was associated with multiple episodes of vomiting. Upon admission, the patient was alert and oriented. His vital signs were a BP of 138/100 mmHg, a pulse of 90, a temperature of 36.4 °C, a respiratory rate of 20, and a SpO2 of 98 %. Physical examination revealed tenderness in the epigastric area, while the rest of the examination

was benign. The patient was fully vaccinated and had no significant medical or familial history. Laboratory findings upon admission were as follows: the WBC count was high at 22.21, with a predominance of neutrophils (85.90 %) and a decrease in the lymphocyte count (10 %). His hemoglobin level was high at 15.50 mg/dL, RBC count was high at 5.52*1012/L, and platelet count was 244,000 µL. The patient's MCV was low at 80.60 fL, MCHC was high at 34.8 g/dL, MPV was high at 9.80 fL, and cGlu-ven was high at 6.9 mmol/L.

The patient underwent a C-X ray. Multislice computed tomography (MDCT) of the abdomen and pelvis with intravenous contrast revealed a distended stomach with abnormal orientation, a normal gastroesophageal junction position, twisting and narrowing with surrounding fat stranding at the gastroesophageal junction and antrum, resulting in gastric volvulus. There was also left diaphragmatic eventration with herniation of the spleen and left kidney into the chest, and small bowel loops were pushed to the right side of the abdomen. There was an organo-axial volvulus of the stomach and decreased wall enhancement of the gastric wall, suggesting impending gastric ischemia and severe compression of the gastric artery. Mild free fluid was observed in the pelvis with no free air (Fig. 1). The patient was prescribed cefuroxime sodium (750 mg), dextrose 5 % and NaCl 0.9 % solution (2400 mL), and pantoprazole (40 mg) for abdominal pain and vomiting.

1. Download: Download high-res image (179KB)
2. Download: Download full-size image

Fig. 1. Computed tomography scan (CT) of chest, abdomen, and pelvis showing a large Bochdalek's Hernia containing spleen and kidney.

Surgery was performed with a midline incision in the abdomen. The peritoneal cavity was reached, and the stomach was gently derotated. A Bochdalek hernia was identified, with the herina sac exceeding 25 cm in diameter and a diaphragmatic defect measuring 12 cm. The kidney and spleen were reduced back to the abdominal cavity, and the hernia sac was dissected and removed. The diaphragm was repaired primarily using 5 U-shaped stitches with no 2 Polyester sutures (Fig. 2). The stomach was fixed to the anterior abdominal wall using Polyglactin 910 sutures at 3 sites: the right hypochondrium, left hypochondrium, and epigastric region. The abdomen was closed, and the patient was transferred to the recovery room. A few days after surgery, his inflammatory markers improved, he had no fever or abdominal pain, and the wound site was clean and dry. Three days after the operation, a chest X-ray revealed left-sided hydropneumothorax, pneumothorax 27 mm from the apex, left-sided base atelectasis, no significant shift of the mediastinum, and no signs of tension pneumothorax, and the left hemidiaphragm was not well outlined due to pleural effusion (Fig. 3). An 8 French pigtail catheter was inserted into the left pleural cavity, and bloody fluid was aspirated. On day 8 after the operation, the patient's status was stable, and his vital signs and tests, including RBC, Hgb, CRP, and PCT, were within normal ranges despite a slightly elevated WBC count of $10.01 \times 10^9$/L. A left-sided pigtail drain with suction drained 200 mL of milky yellow fluid over 24 h. On day 10, a follow-up chest X-ray revealed minimal basal congestion in the left lower zone and the absence of pneumothorax. The pigtail drain had drainage of 75 mL of serous fluid in the last 24 h without suction; therefore, it was removed 2 days later. The patient was discharged 11 days after the operation.

1. Download: Download high-res image (170KB)

2. Download: Download full-size image

Fig. 2. The hernia defect post-repair.

1. Download: Download high-res image (83KB)
2. Download: Download full-size image

Fig. 3. X-ray of the chest showed left-sided hydropneumothorax.

## 3. Discussion

BH is a congenital abnormality located in the posterior lateral diaphragm that allows the migration of abdominal viscera into the thorax. This condition results from the failure of the pleuroperitoneal ducts to close during the eighth week of gestation [7,8]. BH affects approximately 1 in 2200–2500 live births and has a male-to-female ratio of 3:1 [8]. The majority of cases, approximately 80 %, occur on the left side, while right-sided hernias are less common, accounting for approximately 10 % of cases [7,8]. Left-sided hernias may be more likely to cause symptoms than right-sided hernias [7]. In a literature review by Brown et al., precipitating factors or inciting events were identified in 25 % of adult BH patients. Pregnancy was found to be the most common and dangerous factor, contributing to increased intra-abdominal pressure. Other factors included obesity, trauma, constipation, coughing, and chronic obstructive pulmonary disease (COPD) [7]. In the case we examined, the patient had no previous medical history and did not report any initiating incidents or triggering factors. BH in adults can present with a variety of abdominal symptoms, such as abdominal pain, vomiting, and dysphagia, or chest symptoms, such as chest pain, pleural effusion, dyspnea, and recurrent lower respiratory tract infections. In some instances, it can remain asymptomatic for years until the hernia causes respiratory or abdominal symptoms or is discovered incidentally [[8], [9], [10]]. In our case, the patient presented with diffuse abdominal pain, tenderness in the epigastric region, and recurrent vomiting but did not exhibit any chest symptoms. The diagnosis of BH can be challenging due to its rarity and the variety of symptoms it presents [1]. Often, it is diagnosed incidentally via a computed tomography (CT) scan, which is considered the gold standard technique for diagnosing BH [7]. Our patient underwent an urgent pelvic and abdominal CT scan to rule out appendicitis. The scan revealed a hernia on the left side and a gastric volvulus, leading to a quick and definitive diagnosis.

Incorrect diagnoses can lead to inappropriate procedures, such as the placement of a chest tube [7]. In a review study by Thomas et al., 38 % of 51 adult patients diagnosed with BH were found to have received an incorrect diagnosis [11]. Patients who do not undergo a CT scan before surgery could be misdiagnosed with conditions such as pneumothorax, empyema, pleural effusion, or lung cysts. A lack of awareness during the examination could also result in a false diagnosis of conditions such as intestinal obstruction, diaphragmatic tumor, subphrenic abscess, pancreatitis, mediastinal tumor, hiatus hernia, or diaphragmatic eventration [11]. The size of a hernia is determined by its contents, which can vary among patients [7]. In our case the hernial sac was huge, exceeding 25 cm in diameter, and the diaphragmatic defect was 12 cm. Most of the time, the peritoneal sac is absent [12]. When present, it may contain multiple viscera, such as the small intestine, colon, stomach, and spleen [8]. In rare cases, it can contain the pancreas, appendix, ovary, and Meckel's diverticulum [12]. In our patient, the hernia sac contained the spleen and left kidney. Congenital abnormalities, such as an ectopic kidney, may be present in adults with BH. Of all the locations of ectopic kidneys, the intrathoracic position is the rarest, with a reported prevalence of 5 % [4]. The incidence of BH related

to ectopic kidneys is reported to be less than 0.25 % [4]. This disorder predominantly affects men, with 80–90 % of cases occurring in males, and is more frequent on the left side [4]. Most ectopic kidneys and BH are identified accidentally, for example, during surgery or an X-ray for another medical condition in a child or adult [4]. According to a systematic review of intrathoracic kidney patients from 1988 to 2018, 85 % of the patients did not experience any complications. The reported complications included pyelonephritis, hydronephrosis, renal calculi, renal artery stenosis, and cancer [5]. Notably, in our patient, the patient exhibited no clinically or radiologically evident kidney complications. An extension or absence of gastrosplenic and gastrophrenic ligaments in congenital diaphragmatic hernia may predispose patients to stomach volvulus due to the existence of a diaphragmatic defect [12]. A gastric volvulus is described as a twisting of the stomach that results in a closed-loop obstruction, typically requiring 180° of torsion to produce such an obstruction [12]. Gastric volvulus is considered a rare condition that is primarily caused by Hiatus hernia [13]. If left undiagnosed or untreated, serious consequences could occur, including perforation in the stomach, gastric ischemia, necrosis, and sepsis, which could increase the mortality rate [13]. Therefore, gastric volvulus is an emergency case that requires immediate intervention. In this study, we reported a case of gastric strangulation with severe compression of the gastric artery resulting from gastric volvulus. Surgical repair is recommended for adult patients diagnosed with BH to prevent the risk of life-threatening complications, including hernia incarceration, obstruction, strangulation, and perforation [3]. The mortality rate for surgical intervention for early-diagnosed BH is 10 %. However, the incidence of this condition increases to between 20 % and 80 % for emergency surgeries due to strangulated or perforated bowel [14]. Our patient underwent urgent surgical intervention due to significant compression of the gastric artery leading to impending gastric ischemia. Numerous studies have outlined the effectiveness of thoracoscopic and/or laparoscopic BH repair in adults; however, surgical intervention remains the definitive treatment [1,3]. There are several surgical techniques documented to manage BH, including abdominal, thoracic, or thoracoabdominal approaches. Among these, the abdominal approach is the most widely adopted [3]. In this case, we used the abdominal method to reach the hernia with a midline incision. The surgical procedure includes closing the hernia opening and repositioning the protruding organs [15]. Surgical correction of hernias linked to intrathoracic kidneys has been documented without any notable complications [5]. In our case, the surgery included derotation of the stomach, reduction of the spleen and kidney into the abdominal cavity, and removal of the hernia sac. The mortality rate in adults is less than 4 % [11]. Postoperative complications, though infrequent, include persistent pneumothorax, pleural effusion, vena cava obstruction, intestinal obstruction, and lung hypoplasia [11,15]. In our patient, we observed hydropneumothorax 2 days after the operation, which improved during the follow-up period.

## 4. Conclusion

This case report underscores the importance of considering Bochdalek's hernia (BH) in adults who present with abdominal and chest symptoms. Despite its rarity, BH can lead to severe complications such as gastric volvulus and strangulation, necessitating prompt diagnosis and surgical intervention. This case contributes to the limited literature on BH in adults and emphasizes the need for further research in this area.

## Abbreviations

BH

Bochdalek's hernia

Abstract

We report a case of a 65-year-old male, who presented with respiratory complaints of cough and breathlessness, managed initially as respiratory tract infection. However, the patient did not improve, and a thorough examination and imaging revealed herniation of a gut segment into the thorax. The patient was operated and respiratory symptoms improved dramatically.

1. Case

A 65-year-old male ex-smoker, farmer by occupation, presented with a history of dry cough and progressive breathlessness, of two weeks duration. There was no similar history in the past. The patient did not have any history of trauma, fever, haemoptysis, constipation, abdominal or chest pain.

On examination, the patient was conscious, cooperative, oriented but breathless. He was of an average built with BMI of around 25.8 kg/m$^2$. He was tachycardic (pulse = 110) and had tachypnea (RR = 24). There was no pedal edema or raised JVP. Chest examination revealed decreased movements, a dull note posteriorly with a tympanic note anteriorly and absent breath sounds in right infrascapular region. Bowel sounds were heard in the right infraclavicular area. Abdominal examination was normal. Blood gas analysis showed hypoxemia (pao2 = 54 mm Hg) with mild hyperlactemia (lact = 2.5 mmol/L). Chest X-ray revealed a right lower zone haziness with air levels. Ultrasonography of abdomen was unremarkable. Septic screen and tubercular profile were negative. The patient was treated empirically with antibiotics for community-acquired pneumonia. However, the patient did not improve, and a CT chest was done that revealed herniation of bowel loops into the chest through a right anterior diaphragmatic defect. Subsequently, the patient was managed surgically.

1. Download: Download high-res image (489KB)
2. Download: Download full-size image

2. Discussion

MH was first described by Giovanni Battista Morgagni, an Italian anatomist and pathologist in 1769, while performing a postmortem examination on a patient who died of a head injury [1]. MH is the most rare of the four types of congenital diaphragmatic hernia (2%–3% of all cases) [2,3]. In adults, it can occur on either side of the sternum through a muscle-free triangular space called the Larrey space, although it is more common on the right. In rare cases, the hernia can be bilateral. Symptomatic adult cases of MH are rarer with only 12 cases described [4]. Most hernias of Morgagni are diagnosed late because patients can be asymptomatic or present with vague gastrointestinal and respiratory symptoms and signs [5]. Men present earlier in life than women. Very few adult patients present with chest symptoms, the majority describing abdominal pain due to strangulation of the viscera[4]. MH usually presents with recurrent chest infections in children (55%) and lateral chest radiographs are usually always conclusive [6]. Screening may apply to children with increased risk associated anomalies and familial forms of congenital diaphragmatic hernias (from 34% to 50%) [7]. Patients with Down's syndrome have increased risk of MH [8]. Obese patients may develop it later in life and sometimes it may follow trauma. Differential diagnosis would be an intrathoracic tumor, atelectasis, pneumonia, or pericardial cyst. Depending on the contents of the hernia—omentum, stomach, small intestine, or liver [9].

Pre-natal diagnosis by ultrasonography is possible in 50 percent to 90 percent of cases [10,11]. The intestine and the liver may be in the thorax and the lungs are small. Ultrasound scans allow detailed

assessment of the heart. Lung growth is measured as a proportion of head growth. The lung-to-head ratio (LHR) has some prognostic value [[12], [13], [14], [15], [16], [17], [18]], because when it is below 1, survival is compromised [19]. After birth, a diagnosis can readily be made on the basis of symptoms and physical signs. Blood gases and pH status reflect the efficiency of gas exchange. An X-ray of thorax and abdomen may be sufficient, but passing a naso-gastric catheter into the stomach before a plain X-ray of the thorax and abdomen may help to locate it or to detect any esophageal displacement [20] . In some rare cases, herniation of viscera through the diaphragm is an incidental finding in adult patients. Computed tomography can be considered to be an accurate, non-invasive method of diagnosing MH. It can help establish a diagnosis if, as in some cases, the hernia sac is empty or contains omentum or part of the liver. But as described by Fagelman et al. the computed tomogram did not confirm the diagnosis after the chest radiograph as the presence of gas within the lesion was variable: the bowel was sliding in and out of the defect [21]. This might make diagnosis difficult or confusing. Other investigations such as magnetic resonance imaging (MRI) and radio nucleotide liver scan may help with diagnosis but the cost is difficult to justify. Collie et al. demonstrated with MRI a herniation of liver through MH on a patient who presented with increasing shortness of breath and exertional angina [22].

When diagnosis is made *in utero*, amniocentesis is often performed for detecting chromosomal aberrations [23] and may help to estimate lung maturity [24]. Surgery is appropriate for the management of symptomatic adult patients with MH, particularly those with findings of intestinal strangulation, with laparoscopic treatment an alternative approach in selected cases.

## 3. Conclusion

MH is rare in both adults and children. Diagnosis becomes difficult when abdominal symptoms are absent particularly in elderly. Diagnosis should be considered always in a patient who has dissemblance in examination and imaging features and in those who do not respond to medical management as treatment of this condition is essentially surgical.


## Background

Chest wall herniation has been described after thoracotomy, trauma, and violent coughing episodes. Few studies have examined risk factors associated with chest wall herniation or predictors of complications after surgical repair.

## Methods

A divisional database identified all patients who underwent chest wall herniorrhaphy between 1992 and 2011. Data were collected on patient age, sex, body mass index (BMI), cause and location of hernia, comorbidities, duration and technique of herniorrhaphy, postoperative complications, and hospital length of stay. Risk factors for chest wall herniation were then examined, and primary repair was compared with prosthetic repair for differences in postoperative morbidity.

## Results

Twenty-seven consecutive patients underwent chest wall herniorrhaphy. Hernias most commonly occurred on the right side, in the fifth intercostal space, contained lung, and were chronic in nature. Pain was the presenting symptom in all but 4 patients. The most frequently observed comorbidities were obesity, chronic obstructive pulmonary disease (COPD), oral steroid use, and diabetes mellitus. Primary repair was performed in 18 patients and mesh repair in 9 patients, with a median operative time of 116 minutes. Excluding the 4 acute hernias repaired during the same admission as the initial thoracotomy, postoperative complications occurred in 22% of patients who

underwent prosthetic repair and 42% of patients who underwent primary repair ($p$ = 0.4). Median hospital stay did not differ between herniorrhaphy techniques.

Conclusions

Previous thoracotomy, obesity, COPD, steroid use, and diabetes mellitus are common in patients in whom chest wall hernias develop. Prosthetic herniorrhaphy is not associated with an increased risk of postoperative complications relative to primary repair.

Patients and Methods

After institutional review board approval was obtained and the need for individual consent was waived, data were collected on all adults who underwent operation for the diagnosis of chest wall hernia at Mayo Clinic between January 1992 and December 2011. Patients were identified using the general thoracic surgery divisional database. Variables not contained in the database were retrospectively abstracted from the medical record. Exclusion criteria included age less than 18 years.

Data were

Results

Twenty-seven consecutive patients underwent chest wall herniorrhaphy during the study period (4 acute cases, 23 chronic cases). Hernias most commonly occurred on the right side, in the fifth intercostal space, contained lung tissue only, and were chronic in nature (median duration, 6 months; range, 5 days–83 months). All patients were symptomatic, with pain being the predominant complaint in all but 4 patients. The most frequently observed comorbidities were obesity, COPD, oral steroid use, and

Comment

The chest wall hernia was initially reported by Roland in 1499 [1]. Since then, only a few small series 2, 3 and case reports 4, 5, 6, 7, 8, 9, 10, 11 have been published describing this condition. In the 19th century, Morel-Lavallee classified lung hernias by location and cause [12]. Because of its rarity, no study has attempted to identify comorbidities associated with acquired chest wall herniation or to compare postoperative complications using various repair techniques.

Abstract

Congenital diaphragmatic hernia (CDH) is a developmental disorder in which the diaphragm, the muscle that separates the chest from the abdomen, does not close during prenatal development, allowing abdominal organs to herniate into the chest cavity. It occurs mainly on the left side (80%-85% of cases). CDH is often identified during prenatal assessment. However, instances of late-presenting CDH beyond infancy are exceedingly uncommon, contributing to frequent misdiagnosis and delayed therapeutic intervention. We present a case of a 10-month-old female with an uneventful antenatal and perinatal history who presented with respiratory distress and multiple episodes of vomiting. Her vital signs were stable upon arrival, but she was sent to the PICU due to hypoactivity, reduced oral intake, and agitation. After an urgent CT scan, a herniation of the small and large bowel loops into the right hemithorax was discovered, along with a defect in the right hemidiaphragm. This resulted in a pleural effusion on the right side, a partially collapsed left lung, and a mediastinal shift to the left. The diaphragmatic hernia was corrected through a lateral thoracotomy at the sixth rib with multiple interrupted sutures, and a chest tube was then inserted into the pleural space above the diaphragm following a smooth reduction of the bowl. This case

highlights the importance of early diagnosis, appropriate clinical investigation, and treatment. A good prognosis can be anticipated by promptly discovering and examining the condition.

## Introduction

Congenital diaphragmatic hernia (CDH) occurs due to a defect in the incomplete muscularization of the diaphragm. Usually, this condition occurs in the neonatal period, presents with respiratory distress symptoms, and could be associated with pulmonary hypoplasia. The late-presenting CDH, however, is relatively infrequent and usually occurs in later childhood, consisting of vague gastrointestinal or respiratory symptoms [1]. The incidence of reported late-presenting CDH varies from 5% to 45.5% of all cases of CDH [2,3]. Diagnosing CDH in the childhood period can be challenging due to the nonspecific broad spectrum of symptoms that may vary from mild to moderate gastroesophageal reflux and respiratory distress that does not respond to medical therapy or as an incidental finding on a routine chest X-ray done for suspected respiratory infection [4]. We are presenting here a case of a 10-month-old female patient with a history of nonspecific respiratory symptoms whose final diagnosis was a right-sided diaphragmatic hernia. This case report highlights the importance of familiarizing with the natural history of this condition in childhood and underscores the course of management compared to neonatal CDH.

## Presentation

This 10-month-old girl came to the ER complaining of respiratory distress and vomiting that is not bloody, not gelatinous, and not projectile, with decreased oral intake and suboptimal activity, all of which started today. No trauma, no rash, no diarrhea, no fever, no runny nose, no altered level of consciousness, and no signs of child abuse.

On inspection, the child was hypoactive, stressed, and slightly dehydrated, so he was admitted to the pediatric intensive care unit.

Examination of the child reveals good tone and posture. Auscultation of the chest shows decreased air entry on the right side, without crepitation or wheezing. The abdomen is soft and without HSM or masses.

Vital signs: rectal temperature 36.7, O2 saturation 95% on room air, blood pressure 100/60, heart rate 120, and weight 8.9 kg.

## Labs

ABGs before fluid administration: PH = 7.2, H = 12, after resuscitation: PH = 7.3, H = 18.

The lactic acid value was ER 4.5; after the fluid, it was lowered to 2.2.

Abdominal X-ray and chest X-ray (Fig. 4, Fig. 5) were performed and revealed either a diaphragmatic event or a hernia.

The abdominal ultrasound image shows that the liver has been pushed to the left side, mainly through herniated intestinal loops into the right side of the chest. The liver, spleen, gallbladder, and both kidneys are largely normal. The urinary bladder is partially filled, and there is no free fluid.

A CT scan revealed a diaphragmatic hernia with small bowel contents in the right chest (Fig. 1, Fig. 2, Fig. 3). Therefore, the patient was scheduled for an urgent surgical reduction and repair of the hernia, (Fig. 6, Fig. 7, Fig. 8) shows X-rays of the patient chest and abdomen immediately after the surgical repair, postoperative day 1, and before the discharge respectively.

1. Download: Download high-res image (319KB)
2. Download: Download full-size image

Fig. 1. Coronal CT shows herniated bowel loops.

1. Download: Download high-res image (288KB)
2. Download: Download full-size image

Fig. 2. Coronal CT shows diaphragmatic defect.

1. Download: Download high-res image (170KB)
2. Download: Download full-size image

Fig. 3. Axial CT shows herniated bowel loops.

1. Download: Download high-res image (324KB)
2. Download: Download full-size image

Fig. 4. Fluid in the right chest and mediastinum shift to the left side.

1. Download: Download high-res image (325KB)
2. Download: Download full-size image

Fig. 5. Opacification in the right chest and mediastinum shift to the left side.

1. Download: Download high-res image (548KB)
2. Download: Download full-size image

Fig. 6. Immediate postoperative X-ray.

1. Download: Download high-res image (424KB)
2. Download: Download full-size image

Fig. 7. Postoperative day 1 X-ray.

1. Download: Download high-res image (523KB)
2. Download: Download full-size image

Fig. 8. X-ray just before patient discharge.

## Discussion

This case illustrates the challenges associated with diagnosing late-presenting congenital diaphragmatic hernia (CDH). Congenital diaphragmatic hernia (CDH) is a well-recognized entity occurring in 1 out of 25,000 to 30,000 babies [5]. Incidence is around 1 in every 30,000 live births, male-to-female ratio was approximately 2: 1 [6,3]. The incidence of late diagnosis is variable, ranging from 3% to 5% [7].

It is an acquired herniation of the abdominal viscera that occurs through a congenital defect that the spleen or liver had occluded [8,9]. Beyond the neonatal period is not uncommon.

In our case, a 10-month-old female presented with respiratory distress and vomiting, symptoms that initially led to a broad differential diagnosis. Its diagnosis in infancy and early childhood poses a challenge owing to different clinical presentations ranging from gastrointestinal to respiratory symptoms. These neonates are usually misdiagnosed as having pneumonia until radiological imaging picks up the defect during routine scans for worsening respiratory symptoms [10].

There is a high association of pulmonary hypoplasia and pulmonary hypertension. The degree of lung hypoplasia and pulmonary hypertension are key determinants of the morbidity and mortality of these patients [11]. The commonest 2 types of foramina where the hernia can occur are Bochdalek and Morgagni. They can be unilateral or bilateral.

The patient's initial evaluation, including a chest X-ray and abdominal ultrasound, suggested a diaphragmatic event or hernia. The diagnosis was confirmed by a CT scan, which revealed herniation of the small bowel into the right hemithorax and a defect in the right hemidiaphragm.

The most common is the Bochdalek type. The Bochdalek type has a posterolateral position, which can result in a mediastinal shift on the contralateral side and pulmonary hypoplasia on the same side of the hernia [12,13].

Symptoms comprise a wide range from mild to moderate gastroesophageal reflux to respiratory distress [4] after the neonatal period the symptoms may be varied like recurrent vomiting or upper respiratory infections [14,15]. The diagnosis occurs in 50–90 % percent with antenatal ultrasonography [16]. Others diagnosed by a chest X-ray, which will show an air-fluid level, in addition to the contrast material administration by a nasogastric tube which will help in the diagnosis [17]. Although, the chest CT scan is the most sensitive diagnostic method for diaphragmatic defects [18].

The surgical approach involved reducing the herniated bowel and repairing the diaphragmatic defect, most patients with late-onset congenital diaphragmatic hernia will treated and do well with a surgical repair [19]. A diaphragmatic hernia may lead to acute gastric volvulus [20], in addition to the risk of gastric strangulation, ischemia, perforation, pancreatitis, peritonitis, shock, and death with a mortality of 80% [20,21].

This case underscores the importance of considering CDH in the differential diagnosis of infants with unexplained respiratory symptoms, as early diagnosis and intervention can significantly improve outcomes.

In conclusion, this case emphasizes the importance of maintaining a high index of suspicion for CDH in infants with atypical respiratory or gastrointestinal symptoms. Early and accurate diagnosis, supported by appropriate imaging, is crucial for timely surgical intervention and favorable outcomes.

Future research should focus on improving diagnostic strategies and understanding long-term outcomes in late-presenting CDH cases to enhance clinical management and prognosis.

Conclusion

This instance underscores the diagnostic difficulties and crucial management of late-presenting congenital diaphragmatic hernia (CDH) in a 10-month-old girl with unspecific respiratory symptoms. Although relatively rare, late-onset CDH presents significant diagnostic difficulties due to its varied often nonspecific clinical manifestations that can present as mild gastrointestinal symptoms or respiratory distress mimicking common pediatric conditions such as pneumonia. This case highlights the need for a high index of suspicion for congenital diaphragmatic hernia (CDH) in children with unexplained respiratory problems and the importance of immediate X-ray evaluation for definitive diagnosis.

The successful surgical repair in this patient suggests a favorable prognosis with timely intervention. The need for early detection to prevent potential complications like gastric volvulus, strangulation, and other severe morbidities related to untreated CDH was also emphasized. Physicians should be familiar with the natural history and presentation complexity of CDH beyond the neonatal period so that prompt diagnosis and treatment can be made. Subsequent research should focus on improving diagnostic approaches and determining long-term outcomes among those suffering from late presenting CDH cases thus further enhancing clinical management and prognosis.

Abstract

Introduction and importance

Congenital diaphragmatic hernia (CDH) is rare, occurring in 1 in 2000 to 4000 live births, and is typically diagnosed in neonates. Bochdalek hernia is the most common type, usually presenting as a left-sided posterolateral defect. Adult presentations of CDH are uncommon and often incidental. This report discusses a young adult with an undiagnosed CDH, emphasizing the importance of clinical awareness.

Case presentation

A 26-year-old man presented with flu-like symptoms and stable vital signs. He reported chronic postprandial shortness of breath that improved with standing. Physical examination revealed decreased breath sounds on the left side. A chest X-ray identified a left diaphragmatic hernia, confirmed by spiral chest computed tomography. Although advised to undergo surgery, the patient opted for discharge against medical advice.

Clinical discussion

Bochdalek hernia, comprising over 95 % of CDH cases, is usually left-sided due to a defect in the pleuroperitoneal membrane. Adults with CDH often present with nonspecific symptoms or the condition is discovered incidentally. Our patient adapted to his symptoms by standing after meals, which provided relief. Surgical intervention is recommended to prevent organ damage, with various techniques available, including open and endoscopic surgery. This case highlights the necessity of clinical vigilance in diagnosing CDH in adults.

Conclusion

Adult congenital diaphragmatic hernia, though rare, requires prompt surgical treatment to prevent organ damage. Recognizing subtle symptoms is crucial for diagnosis. This report contributes to the

limited literature on adult-diagnosed CDH, stressing the need for clinical awareness and timely management.

## 1. Introduction

Congenital diaphragmatic hernia (CDH) is a rare condition. The incidence of diaphragmatic hernia is based on previously performed epidemiological studies of 1 in 2 to 4000 live births. Although there have been reports of diaphragmatic hernia in many newborns, a diaphragmatic hernia report is scarce in adults [1]. There are several variants, the most common being the Bochdalek hernia, which accounts for approximately 90 % of cases. It is characterized by a posterolateral defect, usually occurring on the left side. Other forms include the Morgagni hernia, which is located anteriorly, and central tendon defects. Rarely, patients can reach adulthood undiagnosed and usually present with respiratory and gastrointestinal symptoms [2]. We present a young man with a congenital diaphragmatic hernia. This study has been reported in line with the SCARE criteria [3].

## 2. Case presentation

We present a 26-year-old man with flu-like symptoms who was referred to the clinic. Written informed consent was obtained from the patient to publish this case report and any accompanying images. His vital signs were stable (No fever, tachypnea, or tachycardia), and his symptoms suggested a common cold. The patient did not have dyspnea and had a dry cough three days before. The patient was lean (Body mass index = 18.2 kg/m$^2$). In the chest examination, a decrease in breath sounds was evident on the left side of the chest. There is no history of thoracoabdominal trauma. The patient admitted that since childhood, he has felt shortness of breath after every meal, which has improved as long as he stands or walks and usually lasts 45 to 90 min. So, the PA chest X-ray was performed. The left diaphragmatic hernia was seen in the chest X-ray (Fig. 1). For more evaluation, a spiral chest computed tomography scan confirmed the initial diagnosis (Fig. 2, Fig. 3). The patient was introduced for further screening and surgery but discharged against medical advice.

1. Download: Download high-res image (92KB)
2. Download: Download full-size image

Fig. 1. Chest X-ray: The defect is evident in the left diaphragm.

1. Download: Download high-res image (417KB)
2. Download: Download full-size image

Fig. 2. Chest computed tomography scan (axial view): The presence of abdominal organs on the left side of the thorax.

1. Download: Download high-res image (237KB)
2. Download: Download full-size image

Fig. 3. Chest computed tomography scan (coronal and sagittal views): The presence of abdominal organs on the left side of the thorax.

## 3. Discussion

Among the congenital anomalies that are followed by a defect in the closure of the pleuroperitoneal canal posterior to the septum transversum, congenital diaphragmatic hernia (CDH) is the most commonly reported abnormality called Bochdalek defect [4]. More than 95 % of congenital diaphragmatic hernias comprise the Bochdalek hernia, mainly on the left side. On the other hand, the term Bochdalek hernia is a mistake because, in 85 % of cases, an actual hernial sac is not created due to the non-formation of the pleura-peritoneal membrane [5,6]. Late symptoms may include chest discomfort, gastrointestinal symptoms, or hernia strain, leading to a diagnosis. On the other hand, the diaphragmatic hernia may be accidentally seen by accidental chest radiography due to another cause [7,8]. In our case, we accidentally found the diaphragmatic hernia in the chest X-ray. An interesting point about our patient was the adaptation of the patient to dyspnea after eating, which he did by standing up, which helped improve his symptoms. In all adult patients with diaphragmatic hernia diagnosis, to prevent damage to organs, intra-abdominal organs should be repositioned, and defecated diaphragm should be repaired. Various techniques are available for surgical repair of diaphragmatic hernia, including open or less invasive endoscopic surgery [9].

The strength of our report lies in its rarity. It contributes valuable insights into the clinical presentation, diagnostic approach, and management considerations of late-diagnosed CDH. However, our report is limited by its retrospective, single-case nature, which restricts its generalizability to broader patient populations. Additionally, the patient's refusal of surgical intervention precludes insights into long-term outcomes and the efficacy of surgical management. Despite these limitations, our case report provides a foundation for future research to elucidate the epidemiology, clinical features, and optimal management strategies of late-diagnosed CDH.

## 4. Adult Bochdalek hernia

### 4.1. Etiology and pathophysiology

Adulthood-diagnosed congenital diaphragmatic hernia (CDH), particularly the Bochdalek hernia, is a serious clinical entity that requires attention due to its implications for patient care and surgical intervention. CDH is a condition characterized by abnormal development of the diaphragm, a part of the lungs, resulting from defects in the septum transversum, pleuroperitoneal folds, body wall, and dorsal mesentery [[10], [11], [12]]. The etiopathogenesis of CDH is unknown, but it was initially believed to be associated with the failure of diaphragmatic closure [10]. The etiology of CDH is unknown in over 70 % of individuals, with some cases showing autosomal recessive, autosomal dominant, and X-linked inheritance patterns [[13], [14], [15]]. Environmental triggers include vitamin A deficiency, exposure to thalidomide or anticonvulsants, mycophenolate mofetil, and allopurinol, which can impair purine biosynthesis [[16], [17], [18]].

### 4.2. Epidemiology, clinical presentation, and diagnosis

The most common type of CHD is the Bochdalek hernia, a posterolateral defect in the diaphragm. The diagnosis in adults is rare, with less than 200 cases reported [19,20]. Common symptoms of adulthood Bochdalek hernia include shortness of breath, food intolerance, gastroesophageal reflux, nausea, vomiting, abdominal cramping, distension, and abdominal pain [20]. A recent systematic review of Bochdalek hernias in the adult population, including 192 patients, revealed most patients were male (50.5 %) with a mean age of 45.41 ± 20.26 years. The most common patient symptoms were abdominal pain (62.0 %) and pulmonary symptoms (41.1 %), with the majority of hernias on the left side (70.7 %). A precipitating factor or inciting event contributed to a patient's presentation in 25 % of cases, including obesity, exertion, trauma, pregnancy, laughter, and more. Pregnancy was

the most common precipitating factor, while exertion was the most common inciting event [21]. Some cases of congenital diaphragmatic hernias in adults are discovered incidentally, often without prior symptoms.

Various diagnostic techniques are used to evaluate adult patients with Bochdalek hernias. Chest X-ray is the most common initial study, frequently followed by computed tomography (CT) scans, which play a critical role in confirming the diagnosis. Upper gastrointestinal (UGI) or barium studies, and contrast enema are also employed in some cases. Esophagogastroduodenoscopy (EGD) and magnetic resonance imaging (MRI) are less commonly used. Specifically, CT scans are crucial for the final diagnosis of right-sided BH in adulthood, while clinical examination or X-rays rarely lead to a definitive diagnosis. MRI also occasionally contributes to the final diagnosis [22,23]. In adult Bochdalek hernias, the most commonly herniated organs include the colon, small bowel, liver, and kidney. There are also occasional cases where other intra-abdominal organs like the gallbladder or pancreas are involved [20,23].

### 4.3. Management

Surgical repair is the primary treatment for adult Bochdalek hernias [22]. The surgical options vary, including abdominal approaches such as laparotomy and laparoscopy, as well as thoracic approaches like thoracotomy and thoracoscopy [[24], [25], [26]]. Some cases have been reported that have utilized robotic-assisted techniques [27,28]. The choice between these approaches depends on the patient's condition and the hernia's characteristics. Laparotomy and laparoscopy are common, while robotic-assisted procedures and thoracoabdominal approaches are less frequent [22,23]. Hernia repairs generally involve either direct diaphragmatic sutures or mesh-augmented closures [[29], [30], [31]]. Some cases may combine mesh with sutures or use clips for additional support [32,33]. Modern surgical advancements have made minimally invasive techniques such as thoracoscopic and laparoscopic repairs increasingly viable. These techniques offer reduced recovery times and less operative trauma. They are particularly useful in managing incarcerated or strangulated hernias. The laparoscopic approach involves strategic patient positioning to manage complications like pneumothorax during the procedure [19,21,23]. The choice of materials for repair—whether absorbable, non-absorbable, or composite mesh—depends on the specific needs of the repair and the surgeon's preference. Bochdalek hernias typically do not have a hernia sac, unlike other diaphragmatic hernias [21]. When present, management of the hernia sac can vary. Some surgeons recommend excising the sac, while others might use postoperative drains [34,35]. The absence of a hernia sac allows direct communication between the pleural and peritoneal spaces, which requires careful intraoperative handling to prevent pneumothorax [36].

In rare instances, non-surgical management is considered, particularly for patients not fit for surgery or those who refuse it [37,38]. This can involve conservative treatment or specific interventions to manage complications, such as percutaneous nephrostomy or ureteral stenting. These approaches are typically used when patients have mild or no symptoms and when surgery poses significant risks.

### 4.4. Outcome and complications

Following surgical treatment for Bochdalek hernias, patients generally had favorable outcomes with a significant number showing no hernia-related complications [21,23]. Common issues included urinary complications like hydronephrosis from herniated organs, which were sometimes resolved post-surgery [39]. Thoracic complications such as abscesses and lung infections occasionally require complex interventions [40,41]. Gastrointestinal problems like bowel ischemia and perforation were less reported but required surgical management [42]. Mortality, though low, occurred due to

pneumonia, sepsis, and complications from bowel perforation [22]. Patients typically reported improved respiratory function post-repair, despite some experiencing prolonged digestive issues. Overall, the prognosis was positive with rare hernia recurrences reported, underscoring the effectiveness of surgical intervention for Bochdalek hernias [22,23,43].

5. Conclusion

In conclusion, adult congenital diaphragmatic hernia, particularly the Bochdalek hernia, presents as a rare clinical entity with diverse symptomatology and diagnostic challenges. Our case report underscores the significance of prompt recognition through imaging, the variability in clinical presentation, and the critical role of surgical intervention in managing this condition. Despite limitations inherent to single-case retrospective studies and the patient's decision against surgery, our findings highlight the need for a multidisciplinary approach, including advanced imaging modalities and tailored surgical techniques, to optimize outcomes. Future research efforts should focus on refining diagnostic criteria, standardizing treatment protocols, and evaluating long-term outcomes to enhance clinical management strategies for adult CDH.

Introduction

Diaphragmatic hernia is most commonly congenital; however, it can also be acquired, most often due to trauma. It is a life-threatening condition resulting in abdominal visceral incarceration and subsequent mortality.

Presentation of case

Our patient was a 27-year-old mother who presented with upper abdominal pain associated with breathlessness. There was no history of trauma. The chest X-ray suggested the diagnosis of a diaphragmatic hernia which was further confirmed on CT. The decision was made to operate on the patient through a laparoscopic approach using single-lung ventilation. A diaphragmatic rent was identified with the incarceration of the stomach and omentum. The rent was widened further which allowed partial reduction of contents and visualization of the left hemithorax which was entirely contaminated. An additional thoracic approach was opted for which enabled reduction of the herniated stomach. A large perforation was present along the greater curvature, which was resected using a linear endo stapler. The diaphragmatic rent was then repaired primarily. Adequate lavage, aspiration, and mopping were performed along with chest tube drainage. The patient remained stable post-operatively.

Discussion

An optimal surgical repair along with sound perioperative care is essential for the successful management of diaphragmatic hernia. A minimally invasive approach avoided extensive open surgery and the complications that would come with it for a young nursing mother.

Conclusion

Such technically challenging cases can be successfully managed with a minimally invasive approach using sound surgical skills and necessary improvisations.

1. Introduction

A diaphragmatic hernia is a protrusion of abdominal contents through a defect within the diaphragm into the corresponding thoracic cavity [1]. It was first reported by Lazarus Riverius during the post-mortem examination of a 24-year-old male [2].

Diaphragmatic hernia is most commonly congenital due to improper fusion of the embryological components forming the diaphragm. Congenital diaphragmatic hernia (CDH) has an incidence of 0.8–5/10,000 live births [3]. CDH interferes with normal lung development resulting in fetal and neonatal complications.

Diaphragmatic hernias may also rarely be acquired, most commonly occurring as a result of blunt trauma with concurrent injuries. The incidence is <1 % in all trauma [4], rising to 3 % in abdominal injuries [5]. The mechanism suggested is a sudden increase in pleuroperitoneal pressure gradient, surpassing the diaphragmatic tensile strength and leading to rupture at areas of potential weakness along embryological fusion points [[6], [7], [8]]. Acquired diaphragmatic hernia (ADH) can also extremely rarely occur iatrogenically or spontaneously, with just a handful of case studies of the same [9]. Sometimes, these injuries may go undetected or untreated and present as a chronic condition where diaphragmatic rents increase in size over time, eventually leading to herniation and subsequent complications.

ADH is a life-threatening condition due to its propensity to cause abdominal visceral incarceration and gastrointestinal strangulation with an overall mortality rate of up to 31 % [6]. Herniation of the stomach, small bowel, mesentery spleen, and pancreas have all been documented [6].

The treatment of ADH is essentially operative, regardless of the presentation. The surgical approach utilized depends on several factors such as the patient's haemodynamic stability, other associated injuries, and the surgeon's expertise. The most common approach remains a formal laparotomy following the principles of operative trauma. In case of a chronic injury, some time may be afforded to allow optimization of the patient and surgery in a semi-elective setting with a minimally invasive approach. Whichever approach is opted for, the principles of surgery remain the same – identification of the defect, reduction of herniated contents, and repair of the diaphragm. As with any hernia, a tension-free repair is essential to prevent failure or recurrence. An optimal surgical repair along with sound perioperative care is fundamental to the successful management of these patients.

The following case report is of an idiopathic incarcerated diaphragmatic hernia with a perforated stomach in a young adult female which was managed by a combined laparoscopic-thoracoscopic approach. Along with the text are the operative videos of the same, showcasing the technical challenges faced and the improvisations made to deal with them.

The following work hereby presented has been reported in line with the SCARE Surgical Case Report criteria [10].

## 2. Presentation of case

Our patient was a 27-year-old lactating mother hailing from rural Kashmir. Her primary complaint was pain in the upper abdomen for the past 5 days, which was moderate in intensity to begin with but had become increasingly severe since the past 1 day, prompting referral to a tertiary care centre. The pain was aggravated with oral intake and associated with nausea and breathlessness.

A thorough history was taken, which revealed little further clues to the diagnosis. There was no history of any trauma. The patient gave birth to a healthy child 6 months back through a normal vaginal delivery and was still in lactational amenorrhea.

At presentation, the patient was conscious, oriented, and hemodynamically stable. She was maintaining an oxygen saturation of 90 % on room air. Chest examination was significant for

decreased breath sounds over the left lower lung zones. Abdominal examination revealed distension and tenderness over the epigastric area.

We then proceeded to investigate the patient. The complete blood picture was normal with no leucocytosis. The biochemistry revealed an elevated serum amylase to the order of 835 U/L, suggesting a provisional diagnosis of acute pancreatitis. Other biochemical parameters were all within normal limits. However, the most striking investigations, and those which turned the diagnosis on its head, were the chest and abdominal X-rays (Fig. 1, Fig. 2) which showed the presence of the gastric air bubble in the left thoracic cavity, suggesting the rare possibility of a diaphragmatic hernia.

1. Download: Download high-res image (288KB)
2. Download: Download full-size image

Fig. 1. Chest X-ray showing the presence of gastric air bubble in the left thorax.

1. Download: Download high-res image (1MB)
2. Download: Download full-size image

Fig. 2. Abdominal X-ray showing the same findings of diaphragmatic hernia.

An ultrasound scan was also done, which revealed left-sided pleural effusion, but no other significant abdominal findings. Following these investigations, a contrast-enhanced CT scan was carried out (Fig. 3). It reported a 25 mm defect in the middle portion of the left diaphragm with herniation of the stomach fundus through the defect into the left hemithorax causing loss of left lung volume along with moderate pleural effusion. The right lung fields and the rest of the abdomen and pelvis were unremarkable for any other findings.

1. Download: Download high-res image (594KB)
2. Download: Download full-size image

Fig. 3. CECT scan demonstrating findings of diaphragmatic hernia.

The pleural fluid analysis showed a polymorphic leucocytosis, and an exudative effusion as per Light's criteria. Serum lipase was normal, and all other causes of pancreatitis were investigated for and ruled out.

With the patient developing a rising trend of leucocytosis and dropping oxygen saturation to below 70 % on room air, the decision was made to operate the patient through a laparoscopic approach using single lung ventilation with the help of a double lumen endotracheal tube. A total of seven ports were utilized – 4 abdominal ports followed by a further 3 thoracic ports (Fig. 4).

1. Download: Download high-res image (457KB)
2. Download: Download full-size image

Fig. 4. Port placement utilized for the surgery.

See Inline Supplementary Video 1

Media player

PlayRestartRewindForward

0 seconds

Volume

SlowerFaster

PreferencesEnter full screen

0:00 / 1:09Speed: 1x*Stopped*

Download: Download video (3MB)

Inline Supplementary Video 1. Introduction to the case

A supraumbilical incision was made and pneumoperitoneum was created using the closed technique. Upon entry into the abdomen, a quick diagnostic laparoscopy was performed to identify the diaphragmatic rent with incarceration of the stomach and omentum. The abdominal working ports were then inserted as shown in the figure above.

Attempts were made to reduce the herniated contents but adhesions were encountered. The rent was further widened using a monopolar energy device followed by an ultrasonic scalpel. On opening up the rent, sero-purulent fluid was found pouring from the thoracic cavity into the abdomen.

The widening of the defect enabled the reduction of the herniated omentum. The scope was then passed through the rent and up into the left hemithorax. The entire left pleural cavity was filled with stomach contents, pus flakes, and infected fluid which was promptly aspirated.

See Inline Supplementary Video 2

Media player

PlayRestartRewindForward

Volume

SlowerFaster

PreferencesEnter full screen

0:00 / 1:00Speed: 1x*Stopped*

Download: Download video (49MB)

Inline Supplementary Video 2. Diagnostic laparascopy and adhesiolysis

The difficulty encountered in reducing the hernia through an abdominal approach made us opt for a thoracic approach. A thoracic optical port was inserted which further revealed the extent of local contamination. Thoracic working ports were inserted and finally, with a combination of traction from above and below, the herniated stomach was reduced into the abdomen.

On inspecting the stomach, a large perforation was found along the greater curvature near the fundus. The anaesthetist was asked to insert a nasogastric tube which was quickly passed along

beyond the perforation into the distal portion of the stomach. Given the extensive size of the perforation, a stapled resection was performed using a 60 mm linear endo stapler.

See Inline Supplementary Video 3

Media player

PlayRestartRewindForward

Volume

SlowerFaster

PreferencesEnter full screen

0:00 / 1:17Speed: 1x*Stopped*

Download: Download video (61MB)

Inline Supplementary Video 3. Thoracoscopy, hernia reduction and stomach resection

Following this, generous lavage and aspiration were performed in the thoracic cavity. We then proceeded to repair the diaphragmatic rent from the thoracic side as it provided better visualization and convenience in suturing. Since the thoracic working ports were only 5 mm in size, the suture was introduced through an abdominal working port and passed into the thorax through the rent. The margins of the defect were freshened and a primary repair was performed using 2–0 barbed delayed absorbable sutures in a continuous manner. The repair was done primarily without the use of a mesh given the extensive contamination present.

See Inline Supplementary Video 4

Media player

PlayRestartRewindForward

Volume

SlowerFaster

PreferencesEnter full screen

0:00 / 1:19Speed: 1x*Stopped*

Download: Download video (51MB)

Inline Supplementary Video 4. Repair of diaphragm

After the repair was complete, the anaesthetist was asked to inflate the left lung which expanded well. Final lavage of the abdomen was done and the specimen was retrieved. Lastly, an intercostal chest tube was placed to facilitate drainage of any residual effusion, along with an abdominal drain.

See Inline Supplementary Video 5

Media player

PlayRestartRewindForward

Volume

SlowerFaster

PreferencesEnter full screen

0:00 / 0:33Speed: 1x*Stopped*

Download: Download video (19MB)

Inline Supplementary Video 5. Lavage and lung expansion

The patient remained stable in the post-operative period, initially requiring intensive care but with quick improvement was shifted to regular inpatient care. The post-operative X-ray is shown (Fig. 5). Following clinical improvement, the nasogastric tube was removed on POD3 and the patient was initiated on clear oral liquids. The patient was weaned off oxygen support and the intercostal chest tube was removed on POD5. The abdominal drain was removed on POD7 and the patient was discharged satisfactorily. Sutures were removed on the first follow-up after a week. The patient remained on regular follow-up, with no further complications.

1. Download: Download high-res image (461KB)
2. Download: Download full-size image

Fig. 5. Post-operative day 1 chest X-ray.

See Inline Supplementary Video 6

Media player

PlayRestartRewindForward

Volume

SlowerFaster

PreferencesEnter full screen

0:00 / 0:41Speed: 1x*Stopped*

Download: Download video (1MB)

Inline Supplementary Video 6. Post-operative course

## 3. Discussion & conclusion

The case report outlines the successful management of an idiopathic incarcerated diaphragmatic hernia with a perforated stomach through a unique combination of laparoscopy and video-assisted thoracoscopy. It yet again highlights the unpredictability of surgery and the adversities posed to a surgeon. However, with sound surgical skills and the incorporation of necessary improvisations, such technically challenging cases can be managed smoothly.

The use of a combined laparoscopic and thoracoscopic approach has been well published in literature primarily for diaphragmatic hernias and esophageal malignancies [[11], [12], [13]] but also for rare conditions such as hepatocellular carcinoma [14], epiphrenic diverticulum [15] and retroperitoneal compound paraganglioma [16].

A combined approach was described for a large traumatic diaphragmatic hernia by Zubaidah et al. in 2015 [17]. It has also been published for congenital diaphragmatic hernia in a child [18], in an adult [19] and with right-sided intrathoracic kidney [20]. Similar to the case described in this report, the use of a combined laparoscopic and thoracoscopic approach in an emergency setting in India was published by Gandhi et al. in 2019 for tension gastrothorax in a foramen of Bochdalek hernia [21].

The use of a minimally invasive approach avoided the formal laparotomy and possible additional thoracotomy which such a case would usually entail, and the complications that would have come with it to a young nursing mother. It allowed for an earlier and easier recovery, minimizing the inevitable post-operative respiratory complications, especially since the patient was already compromised from a respiratory standpoint before surgery. It is to be remembered that such cases not only rely on fine surgical expertise but also anaesthetic proficiency, the unwavering support of whom is invaluable to performing such a procedure.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. Identifying details of the patient have been omitted.

Abstract

Cough is a defense mechanism for airway protection and is associated with multiple systemic complications such as ribs fracture. Diaphragmatic rupture is commonly caused by blunt or penetrating trauma. We presented a case of a 72-year-old female with a 1-year history of chronic cough, not responding to medical management. Imaging showing abdominal herniation into the thoracic cavity and rib fracture due to diaphragmatic and chest wall rupture. Abdominal herniation and diaphragmatic rupture were repaired through surgery allowing resolution of symptoms. This is a life-threatening condition with a high-mortality rate in which early diagnosis and repair are desirable. Therefore, awareness of this uncommon complication of cough should be acknowledged.

Introduction

Cough is a physiologic defense mechanism to protect airways from foreign material and secretions. It is one of the most common symptoms responsible for outpatient clinic evaluation with approximately 30 million visits annually in the United States. It is classified according to the duration as acute (less than 3 weeks), subacute (3-8 weeks), and chronic (more than 8 weeks). Sustained cough can be associated with multiple complications including cardiovascular, gastrointestinal, genitourinary, musculoskeletal, neurologic, ophthalmologic, psychosocial, respiratory, and skin complications [1]. We reported a case with cough-related diaphragmatic rupture which incidence is unknown since condition is rare and many cases likely go undiagnosed.

Case history

A 72-year-old woman G3P3A0 with a medical history of morbid obesity, hypertension, fibromyalgia, diabetes mellitus type 2, and controlled sleep apnea with no toxic habits.

She came to the emergency department with a chief complaint of dry cough and progressive shortness of breath of approximately 1 year of evolution. She mentioned multiple visits to her primary physician as well as to the emergency room without improvement of symptoms during that year. Her treatment included nasal and inhaled steroids, proton pump inhibitors, antibiotics,

and expectorants. As she continued with a prolonged forceful cough not responsive to medical management and developed upper chest pain and a tearing abdominal pain radiated to the back. Chest ecchymosis was present alongside with worsening dyspnea, early satiety upon eating, and left breast pain. She denied fever, sputum production, or recent trauma. Within that year, the patient never got a chest x-ray. Initial chest x-ray (Fig. 1) taken 1 year after the beginning of symptoms showed left-sided diaphragmatic hernia secondary to diaphragmatic and chest wall rupture. Subsequent chest CT scan axial view (Fig. 2), coronal view (Fig. 3), sagittal view (Fig. 4), and reconstruction (Figs. 5 and 6) demonstrated evidence of rib fracture, chest wall, and diaphragmatic rupture with a displacement of small and large bowel into the left side thoracic cavity causing left pulmonary collapse. Surgical findings consisted of a chronic large left anterior diaphragmatic hernia with bowel and omentum protruding to the pleura cavity plus subcutaneous detachment of left subcostal cartilage from the sternum as the causative of lung collapse.
Afterward, thoracotomy was performed with repair of bowel placement on the abdominal cavity, correction of diaphragmatic rupture, reinforcement with a proline mesh, and chest tube placement for pneumothorax. A 1-week postsurgical follow-up with chest x-ray showing resolved herniation (Fig. 7) and discharge without complications.

1. Download: Download high-res image (153KB)
2. Download: Download full-size image

Fig. 1. Initial chest x-ray demonstrates left-sided diaphragmatic hernia secondary to diaphragmatic rupture.

1. Download: Download high-res image (483KB)
2. Download: Download full-size image

Fig. 2. Chest CT with evidence of intra-abdominal content in the left hemithorax. (A) Lung window and (B) abdominal window.

1. Download: Download high-res image (496KB)
2. Download: Download full-size image

Fig. 3. Coronal view of intra-abdominal content in the left-sided hemithorax. (A) Lung window and (B) abdominal window.

1. Download: Download high-res image (216KB)
2. Download: Download full-size image

Fig. 4. Sagittal view with anterior diaphragmatic and chest wall rupture with intra-abdominal content. (Abdominal window.)

1. Download: Download high-res image (287KB)

2. Download: Download full-size image

Fig. 5. Chest CT reconstruction with evidence of a diaphragmatic hernia, rib fractures, and intra-abdominal content of left breast.

1. Download: Download high-res image (227KB)
2. Download: Download full-size image

Fig. 6. Chest CT reconstruction of the lung, remarkable for the left lower lobe collapse.

1. Download: Download high-res image (102KB)
2. Download: Download full-size image

Fig. 7. Chest x-ray postanterior diaphragmatic hernia and chest wall abnormality repair.

## Discussion

Diaphragmatic rupture is most commonly cause by penetrating injury or blunt abdominal trauma in 63% and 37% of the cases, respectively [2]. The diaphragm is a dome-shaped muscle, which contracts during the inspiratory phase. Cough valsalva maneuver causes lack of coordination of different muscles of expiration, the muscle of the abdominal wall contract pushing the diaphragm upward, whereas the ribs are pushed inward and downward leading to a diaphragmatic rupture [3]. Herniation of bowel loops into the chest can be a consequence of diaphragmatic rupture, which impairs ventilation and oxygen delivery. The true incidence of abdominal organ herniation due to diaphragmatic rupture is unknown since many cases likely go undiagnosed or usually appear delayed.

Diaphragmatic injuries are usually diagnostic challenges. Chest radiographs are the initial and most common imaging study to evaluate the diaphragm. When the results are inconclusive, CT is the next study of choice since it can assess the extent and anatomical sites of coexisting thoracoabdominal injuries [4].

The rate of missed diaphragmatic rupture on chest radiographs ranges from 12% to 66% with the potential risk of a late visceral herniation through the diaphragmatic defect [5]. The sum of physician unawareness about the diagnosis, and the subtle and nonspecific findings of chest radiography alongside the technical limitations such as supine positioning, use of portable radiography, and limited patient cooperation make the diagnosis difficult through chest radiography alone. However, it remains a valuable imaging option in the acute phase for the detection of diaphragmatic rupture and when CT is not available or cannot be performed [6,7].

Diaphragmatic rupture has an overall mortality rate of 25% as reported by the National Trauma Data Bank. Due to an increased rate of herniation and strangulation of abdominal organs secondary to diaphragmatic rupture, which can be life threatening, early diagnosis and repair are desirable [8], [9], [10]. Therefore, physicians should be aware of this uncommon complication of cough.

## Introduction

A right side diaphragmatic injury was linked to serious trauma to the abdomen, pelvis, and chest. The most significant type of injury was blunt abdominal trauma sustained in a car collision. The left side

was more likely than the right to experience herniation. The stomach and colon were the most often herniated abdominal viscera. In the same location as the diaphragm rupture, there were rib fractures, hemothorax, and liver damage. Delayed diaphragmatic rupture with diaphragmatic hernia is rare and has a mysterious nature.

Case presentation

A 68 years old female patient who has repeated history of shortness of breath, for which she treated as lung infection presented with sudden exacerbation of shortness of breath, she witnessed history of blunt trauma 20 years back and up on investigation bowel herniation to the chest cavity diagnosed. Posteriolateral thoracotomy done, the herniated bowel reduced and the diaphragmatic defect repaired. The patient significantly improved and discharged from the hospital smoothly on 4th postoperative day.

Discussion

Careful recording of past history and physical examination are the best approaches in diagnosing delayed presentation of traumatic diaphragmatic rupture. CT scan with reconstruction of the diaphragm is helpful in both diagnosis and differential diagnosis. Surgical therapy after diagnosis is the best treatment.

Conclusion

Delayed right side diaphragmatic hernia is a rare entity resulting in grave consequences, In a patient with history of trauma there should be a high index of suspension and patients should undergo imaging and surgical management is the best treatment.

1. Introduction

Traumatic diaphragmatic injury (TDI) is an uncommon but potentially fatal post-traumatic problem. Diaphragmatic ruptures are occurs after thoraco-abdominal traumas (0.8–5 %). In 90 % of cases, they are left-sided. Because the liver often protects the diaphragm, right-sided TDI is particularly rarer. It might be challenging to diagnose and potential for delayed presentation. Although less frequent, right-sided diaphragm hernias can happen and are frequently misdiagnosed .Whatever the situation, TDI must be treated carefully as it might result in bowel strangulation and necessitate immediate surgery. Various methods have been reported for the definitive correction of diaphragmatic abnormalities. We report a traumatic right side diaphragmatic hernia 20 years after she sustained trauma which presented with shortness of breath and successfully managed surgically in low and middle income country and the work has been reported in line with SCARE criteria. [[1], [2], [3], [4]]

2. Case presentation

We present a 68 years old female patient who came from the rural part of the country presented with history of shortness of breath for long duration. For the above compliant she visits several health institutions including our hospital where she was informed that she has respiratory infection and medication was given for that. She was examined with chest X ray four times for same problem and she was told to have pneumonia and treated with antibiotics. The shortness of breath is exacerbated during activity and when she is at rest it usually improves. For this reason she restricted from most of her usual activity and only participated only in home based activity like cooking.

For the last two years the shortness of breath was exacerbated and even starts to limit waking for few distance and she visits private hospital where she again treated as pneumonia but had no

improvement. Over the last 20 years except some intermittent discomfort over the abdomen she had no history of abdominal distension, no history of cramp abdominal pain that necessitates physician visit or no history of failure to pass feaces and flatus.

On her recent visit to our hospital the patient had shortness of breath even at rest with sudden exacerbation of one day duration; otherwise she had no chest tightness, no fever, no cough, no swelling over the extremity or other parts of the body and no history of known chronic medical illness. She has no known familial illness. On further history questioning she gave history of trauma and the mentioned problem came few years after she sustained trauma. She initially denied history of any trauma but later she remembered that she sustained trauma 20 years back where a wall of an old house collapse over right side of chest and abdomen where she visited this hospital and after chest X ray was taken, she sent home with anti-pain. She never taught that her problem is associated with the trauma she sustained while she visits several health facilities for the complaint of her illness. Because of incomplete medical history, lack of imaging modalities and skilled profession in interpreting the clinical condition and the available investigation our patient's problem was not identified on time.

On physical examination, she looks acutely sick looking in cardiorespiratory distress, Her blood pressure 110/70 mmHg; pulse rate 88/m; respiratory rate, 34; temperature,36.1 °C; oxygen saturation, 84 % with atmospheric air and 96 % with Face mask $O_2$. Other physical examination findings she have on chest was decreased air entry over the lower half of the right lung field and audible bowel sound over the right chest and there no pertinent finding on the other system. She was immediately put on face mask oxygen and laboratory examination and chest CT scan ordered.

Her laboratory results were as follows: WBC: 7.5 k/UL with neutrophil: 73.8 %, lymphocyte: 16.6 %, HCT: 38.4 %, PLT: 365 k/uL, Cr:0.7 mg/dl, ESR:5 mm/h. Chest shows bowel in right chest with defect in the diaphragm (Fig. 1, Fig. 2, Fig. 3).

1. Download: Download high-res image (187KB)
2. Download: Download full-size image

Fig. 1. Axial CT scan showing herniated bowel in the chest.

1. Download: Download high-res image (185KB)
2. Download: Download full-size image

Fig. 2. Coronal CT scan showing bowel herniated to the right chest cavity.

1. Download: Download high-res image (178KB)
2. Download: Download full-size image

Fig. 3. Sagittal CT scan image showing bowel in chest through diaphragmatic defect.

The patient was subsequently advised for surgical management after stabilized and further work up was done. After informed and written consent obtained from the patient she undergone right side posteriolateral thoracomy (since the minimal invasive surgery is not available in our set up) and the

intraoperative finding was viable normal size transverse colon in the right which from adhesion with the lung and pericardium compressing the lung (Fig. 4) and there is 3 cm by 4 cm defect on the anteriolateral part of the diaphragm where the neck forms adhesion circumferentially to the herniated bowel. The liver is at its normal site and didn't herniate to the chest and with normal size and length of inferior vena cava which indicates the defect is acquired rather than congenital. There was no fluid or sign of intrapleural infection. The adhesion released gently and the bowel reduced to the abdomen after the neck was widened and the defect in the diaphragm was closed primarily without tension with silk no 2 stitch since the defect is small. Chest cavity closed after chest tube placed (Fig. 5). The post-operative course was smooth and the patients discharged from the hospital on 4th post-operative day with significant improvement and complete lung expansion. The patient is on regular follow up without any post-operative complication and the follow up chest x-ray shows fully expanded lung with on collection in the pleural cavity (Fig. 6).

1. Download: Download high-res image (157KB)
2. Download: Download full-size image

Fig. 4. Intraoperative imaging showing transverse colon in the right pleural cavity.

1. Download: Download high-res image (161KB)
2. Download: Download full-size image

Fig. 5. After the defect primarily repaired.

1. Download: Download high-res image (48KB)
2. Download: Download full-size image

Fig. 6. Follow-up X ray one month after the procedure.

## 3. Discussion

A right side diaphragmatic injury was linked to serious damage to the abdomen, pelvis, and chest. In the same location as the diaphragm rupture, there were rib fractures, hemothorax, and liver damage. Thus, it appeared that the force responsible for diaphragmatic rupture varied between left- and right-sided patients. The blunt blow to the right thoracic wall was the primary cause of the right-sided diaphragmatic rupture. The right lobe of the liver was the most often herniated organ in right-sided diaphragmatic hernias, and delayed presentation of the condition had a significant incidence of G-I tract herniation. The abdominal organ herniation appeared to change over time. Trauma to the thoracoabdominal region is blunt and accounts for 75 % of TDIs. Variable clinical presentations and a high frequency of accompanying life-threatening injuries in blunt TDIs may make it difficult to diagnose and treat the condition early, which might result in more difficult repairs. Diagnostic delays are more probable in cases of right-sided blunt TDIs because they are far less prevalent than left-sided ones, are more difficult to see on imaging examinations, are more commonly linked to other potentially fatal injuries, and tend to present more subtle. It might be particularly difficult to diagnose right-sided TDIs early on [5,6].

Due to the rarity of right side cases, the clinical appearance of a late-diagnosed diaphragmatic hernia necessitates distinct diagnostic and treatment strategies than those for acute diaphragmatic rupture. In patients with a history of trauma, it is important to keep this in mind while making a differential diagnosis. Without a strong index of suspicion and a variable time to diagnosis, traumatic diaphragmatic hernia remains a challenging diagnosis to make. The low frequencies of the illness and its vague clinical presentation have been linked to this. It has been recommended that a chest radiograph be used as the first imaging modality and that computed tomography be used as a reliable adjunct. [[7], [8], [9]]

The sensitivity and specificity of CT-scan for the diagnosis of blunt TDI are both good. When a diaphragmatic hernia is seen on a radiograph or helical CT scan, the diagnosis of blunt TDR can be made with ease. As a preliminary evaluation, “offside sign” is useful for penetrating TDR without hernia. Coronal/sagittal reconstruction on a CT scan is a reliable diagnostic method. [10,11]

Although they are not frequent, surgical repair is necessary for traumatized diaphragmatic injuries (TDIs) in order to treat or prevent herniation. While managing delayed diaphragmatic hernias can be difficult, it is possible to perform a repair that will lead to a satisfactory recovery and a low risk of recurrence with careful planning and an adaptable surgical strategy. An injury that is readily overlooked and can result in considerable morbidity and fatality [7,12].

A minimally invasive (VATS) or an open approach to patients with a delayed-presentation diaphragm hernia is safe and effective. The recommendation for surgical approach is based on patient characteristics, anatomic considerations, and surgeons' experience. Postoperative pulmonary complications including atelectasis are common following surgery for traumatic diaphragmatic hernia. Other complications include surgical infection, bleeding, respiratory failure, ileus, gastroesophageal reflux, chronic pain, hernia recurrence, and cardiac injury [13,14].

4. Conclusion

Post trauma delayed diaphragmatic hernia is rare. As a result, it is important to thoroughly look into the patient's history. Clinical symptoms together with physical findings should be taken into account while making the diagnosis. A reconstruction examination with CT scans of the chest and abdomen can greatly increase diagnostic accuracy. Surgical therapy should begin as soon as the diagnosis is established and before surgery, the surgical strategy and diaphragm repair techniques should be thoroughly assessed in order to have good outcome.

A lung hernia refers to part of a lung pushing through a tear, or bulging through a weak spot, in the chest wall, neck passageway or diaphragm. Lung hernia is a distinctly rare event, regardless of its location and cause. Most lung hernias are acquired traumatic thoracic hernias, most commonly caused by vigorous coughing with a subsequent rib fracture. We report a rare case of lung hernia through the left anterior chest wall in a middle-aged male.  Introduction

Lung hernia is defined as the protrusion of lung tissue covered by parietal and visceral pleurae, beyond the confines of thoracic cavity through an abnormal opening in die thoracic wall. The basic classification of lung hernias was suggested by Morel-Lavellee according to the anatomic location, as cervical, thoracic and diaphragmatic, and according to the etiology, as congenital or acquired. He further differentiated the acquired hernias as traumatic, consecutive, spontaneous and pathologic. The pathologic hernias are secondary to tuberculosis, acute infection or neoplastic diseases of the chest. Most people who experience a lung hernia suffered a severe trauma such as a traffic accident in which the chest is injured. Lung hernias by themselves are not life threatening. They might not

even be all that troublesome. They are fairly easy to locate, diagnose and correct. We here describe a case of old pulmonary tuberculosis with lung hernia through the anterior left chest wall.

## Case report

A middle-aged male presented in the outpatient with complaints of cough with expectoration, dyspnea and dull aching chest pain in the left infraclavicular area. The patient is a known case of old healed koch's. He took antitubercular drugs twice, first almost 11 years back for 12 months with almost 70 streptomycin injections and a second course 6 years back for 10 months. Along with this, the patient also complained of swelling with cough impulse in the left upper chest wall from the last 6 months. This swelling is reducible on applying pressure. There is no history of sudden onset of chest pain or hemoptysis. There was also no history of trauma or surgical intervention on chest. The patient also has history of bidi smoking around 20/day for almost 25 years, which he left 6 months back.

On physical examination the patient was emaciated, anemic and dyspneic. His pulse rate was 98/min, blood pressure 134/68 mmHg and respiratory rate was 30/min. Bronchial breath sounds were heard over the left infraclavicular and mammary areas with inspiratory crepitations over the left mammary area. On local examination, he had ill-defined, smooth, non-tender, soft swelling 9×12 cm in the left infraclavicular and supramammary areas involving I, II and III intercostal spaces, which had a positive cough impulse and could be reduced completely by manual pressure.

The haemogram, urine analysis and blood sugar level were normal. The sputum smear was negative for acid fast bacilli. Spirometry showed obstructive pattern of airflow limitation. The chest roentgenogram, PA view showed fibrocavitary changes involving both upper lung fields along with hyperlucency of both lung fields due to emphysema (Figure 1). CT scan is suggestive of fibrocavitary lesion involving both lung fields with the thin walled peripherally placed cavity herniating through the left upper zone through the intercostal space with surrounding infiltrative changes in the periphery (Figure 2). Thus, a diagnosis of old healed koch's with thoracic lung hernia on the left side was made. The patient was managed conservatively with bed rest, oxygen inhalation, bronchodilators, cough suppressant and analgesics, which relieved him of chest pain and dyspnea.

1. Download: Download full-size image

Figure 1. Chest radiograph showing fibro-cavitary changes involving both the lung fields with emphysematous changes in both the lungs.

1. Download: Download full-size image

Figure 2. CT scan showing fibrotic changes in both lung fields with the thick walled peripherally placed cavity herniating through the left upper zone.

## Discussion

The present case is an example of thoracic lung hernia that, according to the anatomical location, is thoracic as the lung tissue was protruding through the rib cage into the chest wall and, according to the etiology, pathologic as it was secondary to pulmonary tuberculosis.[1] It is known that pulmonary tuberculosis can cause weakness of thoracic wall.[2] Congenital hernia of the lung is due to development defects in the thoracic wall, most frequently in the supraclavicular fossa or anteriorly at

the junction of rib and costal cartilage. Only 1% of the acquired hernias are secondary to tuberculosis, inflammatory or neoplastic disease.[3]

The mechanics associated with lung hernia is as follows: as the person inhales, the lungs expand; this in turn expands the chest; if there is an opening or a soft spot in the chest wall, neck opening or diaphragm, it is possible that the lung as it expands will push through or cause a bulge at that point.[4] Two out of three lung hernias involve the chest (thoracic) wall, and most of the rest involve the neck (cervical) area. Lung hernias related to the diaphragm are uncommon and limited mainly to congenital conditions in infants. In a number of cases, lung hernias occur and persist without bothersome effects. In others, the person might feel sore or have trouble breathing. The development of lung hernia in our patient probably resulted from the fibrocavitary changes associated with old pulmonary koch's along with increase in intrathoracic pressure due to bouts of violent cough.

In mild to moderate cases, a conservative regimen might be sufficient to provide relief, as in our case; the patient was managed conservatively. In the case of a more severe or a larger lung hernia, surgery will be the answer with a periosteal flap from adjacent rib.[5]

Abstract

Hiatal hernia (HH) is a frequent entity. Rarely, it may exert a wide spectrum of clinical presentations mimicking acute cardiovascular events such as angina-like chest pain until manifestations of cardiac compression that can include postprandial syncope, exercise intolerance, respiratory function, recurrent acute heart failure, and hemodynamic collapse. A 69-year-old woman presented to the emergency department complaining of fatigue on exertion, cough, and episodes of restrosternal pain with

The nitrofen model of congenital diaphragmatic hernia (CDH) is widely used in translational research. However, the molecular pathways associated with pulmonary hypoplasia in this model compared to the human CDH phenotype have not been well described. The aim of this study was to investigate differentially expressed genes (DEG) and signaling pathways in early stage fetal lungs in mouse and human CDH.

Methods

CDH lung tissue was obtained from human fetuses (21–23 weeks gestation) and nitrofen mouse pups (E15.5). NovaSeq Flowcell RNA-seq was performed to evaluate differentially expressed transcriptional and molecular pathways (DEGs) in fetal mice with CDH, compared with age-matched normal mouse lungs and human CDH samples.

Results

There were thirteen overlapping DEGs in human and mouse CDH lung samples compared to controls. These genes were involved in extracellular matrix, myogenesis, cilia, and immune modulation pathways. Human CDH was associated with an upregulation of collagen formation and extracellular matrix reorganization whereas mouse CDH was associated with an increase in muscular contraction. The most common cell types upregulated in human and mouse CDH samples were ciliated airway cells.

Conclusions

This study highlights the unique gene transcriptional patterns in early fetal mouse and human lungs with CDH. These data have implications when determining the translational potential of novel therapies in CDH using nitrofen-based animal models.

Level of Evidence

Level IV.

Study Type

Basic science/case series.

Introduction

Congenital diaphragmatic hernia (CDH) is a complex, polygenic disorder characterized by incomplete development of a hemidiaphragm with subsequent intrathoracic abdominal organ herniation. All neonates with CDH have bilateral lung hypoplasia with reduced airway branching and alveolar surface area that is more severe on the side of the hernia defect [1]. Despite ongoing advances in prenatal intervention and postnatal care, mortality rates have been stable at 20–25% over the past several decades. Lack of progress in CDH outcomes can be partially attributed to our incomplete understanding of the underlying molecular mechanisms responsible for abnormal lung morphogenesis.

Although several animal models have been used to study CDH lung development, the most widely used model involves the administration of the herbicide 2,4-dichlorophenyl-p-nitrophenyl ether (nitrofen) to pregnant rodents [2]. Studies have shown that the diaphragmatic defects in nitrofen-exposed fetal mice and rats can have comparable anatomy to the human disease phenotype [3]. However, it has been noted that nitrofen-exposed pups develop lung hypoplasia even in the absence of a diaphragmatic defect [4]. Another major criticism of the nitrofen model is that the teratogen has never been shown to induce CDH in humans. It is therefore unclear the extent to which the molecular pathways implicated in nitrofen-exposed fetal lungs may have translational relevance in the clinical arena.

Here, we applied bulk RNA transcriptome analyses to study hypoplastic lungs from mouse and human CDH samples at a similar stage of fetal development. We specifically sought to compare differentially expressed genes (DEGs) and signaling pathways to understand how the mechanisms behind CDH lung morphogenesis between the two species may be similar or different.

Section snippets

Human and animal samples

Under an approved materials transfer agreement and Institutional Review Board exempt protocol at the Johns Hopkins School of Medicine, canalicular stage human fetal lung samples from isolated CDH at 21–23 weeks' gestation (n = 2) were obtained from the National Institutes of Health Neurobio bank (University of Maryland, Baltimore, MD, USA). Age-matched normal fetal lungs (n = 2) from the biobank were used as controls. Per database regulations, only gestational age and sex were made available;

Results

The rate of diaphragmatic hernia defects in nitrofen-exposed pups was 32% (Fig. 1A). Pulmonary hypoplasia was confirmed at E17.5 based on lung weight adjusted by total body weight (Fig. 1B). In PCA analyses, human CDH and control samples clustered into their respective groups with a PC2

variance of 25% and PC3 variance of 17%. Mouse CDH and control samples at E15.5 had a PC2 variance of 14% and a PC3 variance of 3%. RNA-sequencing in human CDH revealed 493 upregulated and 204 downregulated

Discussion

In this novel study, we compared the transcriptome patterns in both human CDH and murine nitrofen-induced CDH at a similar stage of pulmonary development (early canalicular). Of note, hypoplastic lungs from nitrofen-exposed pups without a diaphragmatic hernia were also analyzed separately, demonstrating an intermediate phenotype similar to control lungs but distinct from lungs with a defect. In view of this

**SESSION TITLE:** Outstanding Thoracic Surgical Cases

**SESSION TYPE:** Rapid Fire Case Reports

**PRESENTED ON:** 10/07/2024 02:35 pm - 03:05 pm

**INTRODUCTION:** Pulmonary herniation through the intercostal space is a very rare complication of epicardial lead placement by thoracotomy. Very few other cases are reported in the literature. We present a case of acute incarceration of a chronic lung hernia resulting from a remote thoracotomy for epicardial lead placement over four years prior.

**CASE PRESENTATION:** A 63 year old woman with a history of heart failure with reduced ejection fraction and left bundle branch block status post cardiac re-syncronization therapy with a defibrillator (CRT-D); coronary artery disease, paroxysmal atrial fibrillation, COPD, and tobacco use presented with shortness of breath, chest pain, and productive cough over the prior three days. Four years earlier, she had undergone surgical removal of an infected implanted cardiac device, with left thoracotomy for epicardial left ventricular lead placement. She had subsequently developed an asymptomatic left lung hernia, in the left 5th to 6th rib interspace, adjacent to the pacemaker lead. In the emergency room, she was found on CT scan to have marked worsening of her lung hernia with ground glass opacities concerning for acute incarceration. Cardiothoracic surgery was consulted, the patient elected to undergo thoracotomy for repair, the hernia was reduced and a 15 cm x 6 cm defect was repaired with mesh. On post-operative day two she developed tachycardia, fever, diaphoresis, and respiratory distress; she was transferred to the ICU for non invasive ventilation and started on broad spectrum antibiotics. Repeat CT demonstrated a dense pneumonia in the left lower lobe. Sputum culture grew Enterobacter cloacae and Haemophilus influenzae. She recovered with a 7 day course of ertapenem.

**DISCUSSION:** This case was unique as few cases of pulmonary herniation exist after epicardial lead placement by thoracotomy. Lung herniation is a rare condition in general, but has been reported after a variety of thoracic surgeries. Risk factors for postoperative lung hernias include conditions resulting in increased intra-thoracic pressure such as COPD, as well as conditions leading to poor wound healing or tissue weakness such as diabetes or malnutrition. Many are asymptomatic, but typical symptoms include dyspnea and chest pain which is often exacerbated by cough. Surgical intervention is recommended for symptomatic lung hernias. In this case, the hernia had been stable for over four years, with imaging as recent as 2 months prior, before the patient developed both symptoms and marked radiologic progression necessitating surgical intervention. There is also an established risk of infection in the post-operative period. Although in this case pneumonia did develop, rapid identification and treatment of infection resulted in a good outcome.

**CONCLUSIONS:** Lung hernia after epicardial lead placement by thoracotomyis a rare occurrence. Even stable, asymptomatic lung hernias must be monitored over time for development of complications and progression. In the post-operative period after definitive surgical management, surveillance for infections is important.

A *lung hernia* is a rare and potentially severe complication that may occur due to thoracic surgery amongst other etiologies. This case report describes the clinical presentation, imaging findings, and management of a patient who developed an iatrogenic lung hernia after undergoing thoracic fusion surgery at the level of T6-T7. The patient presented with persistent chest pain, shortness of breath, and a nonproductive cough. Initial imaging studies revealed the presence of an abnormality within the pleural space, later confirmed through computed tomography of the chest. This case highlights the importance of considering iatrogenic lung hernia as a potential complication of thoracic fusion surgery and the need for close monitoring and prompt intervention in cases when it occurs.  Introduction

Pulmonary hernias are rare and can be caused by various underlying conditions, including congenital abnormalities, conditions that cause chest wall weakening, and traumatic or surgical injury. *Iatrogenic lung hernias* are a rare and often underdiagnosed complication that can occur after thoracic surgery. It results from the herniation of lung tissue through a defect in the chest wall created during the surgical procedure symptoms include persistent chest pain, shortness of breath, and cough, which can often be mistaken for other conditions leading to a delay in diagnosis and treatment. This case report aims to emphasize the importance of considering iatrogenic lung hernias as a potential complication in patients with a history of thoracic surgery, to highlight the computed tomography (CT) imaging findings, and to provide insight into the diagnostic and therapeutic approaches that may be necessary in such cases.

Case presentation

Our patient is a 62-year-old female with a medical history of hypertension and chronic obstructive pulmonary disease (COPD). The patient presented with a chief complaint of severe left-sided chest pain associated with elevated blood pressure, cough, and dyspnea. She had been experiencing less severe pain at the exact location for the past month; however, she reports acute worsening in the severity in the last 3 days. She denied any history of trauma to her chest. In the emergency department, patient vitals were significant for a heart rate of 110 beats per minute, 20 respirations per minute, and blood pressure of 150/86. Laboratory results were within normal limits, including a complete blood count and complete metabolic panel (CBC and CMP). The patient's electrocardiogram and high-sensitivity troponins were all normal x3. The patient's pain subsided with the administration of intravenous morphine.

Her social history revealed 60 pack-years of tobacco use. She also had a surgical history of cholecystectomy and a T6-7 left lateral thoracic fusion performed via thoracotomy approximately 2 years before presentation. Physical examination was significant for increased body habitus, which was unremarkable, including a chest exam.

On admission, the patient's chest X-ray was significant for a “left basilar parenchymal opacification,” portrayed in Fig. 1. This finding was subtle and due to the lack of clarity, a CT chest with contrast was obtained, which uncovered a 4 × 2 cm lung herniation in the lateral seventh intercostal space. Atelectasis associated with the chest wall defect was the reason for the abnormality in the initial chest X-ray. Fig. 2, Fig. 3 portray the radiological CT findings. These findings both explained the

patient's presentation and symptoms. The iatrogenicity of her lung hernia was attributed to the weakened chest wall due to her prior thoracic fusion via thoracotomy.

1. Download: Download high-res image (162KB)
2. Download: Download full-size image

Fig. 1. Patient's initial anterior-posterior X-ray of chest. Black arrow pointing at parenchymal opacity.

1. Download: Download high-res image (222KB)
2. Download: Download full-size image

Fig. 2. Patient's CT scan of chest with contrast, coronal view. Black arrow pointing at lung herniation.

1. Download: Download high-res image (269KB)
2. Download: Download full-size image

Fig. 3. Patient's CT scan of chest with contrast, axial view. Black arrow pointing at lung herniation.

Our patient was informed and counseled on the diagnosis and proposed management. Given the nonacute nature of her lung herniation, symptomatic improvement, and lack of critical signs such as hemoptysis or respiratory failure, the patient was referred to cardiothoracic surgery for outpatient surgical intervention.

Discussion

There are very few cases reported of lung hernias as they are a rare occurrence, Mirza et al. [1] published that less than 300 cases exist in the literature. Although it is suspected that the incidence may be higher than reported, given that some patients may have no symptoms. Consequently, there needs to be more evidence-based guidance for effective management. Iatrogenic lung hernia is a rare but potentially severe complication of thoracic surgery. The herniation of lung tissue occurs through a defect in the chest wall created during the surgical procedure [2]. Diagnosing iatrogenic lung hernia can be challenging, as persistent chest pain, shortness of breath, and coughing are nonspecific and may be mistaken for other conditions, such as in our patient. Early recognition and prompt treatment are essential to prevent serious complications, such as bronchial obstruction, infection, or lung strangulation [3].

The diagnosis of iatrogenic lung hernia is typically made through imaging studies, such as chest X-ray, CT scan, or magnetic resonance imaging (MRI). CT scans are particularly useful in detecting small hernias and assessing the extent of the hernia, while MRI may be more useful in evaluating soft tissue injuries [3]. In some cases, surgical intervention may be necessary to confirm the diagnosis and repair the hernia. The current system used for the classification of lung hernias was first proposed in 1845 and divides lung herniations by location (cervical, thoracic, or diaphragmatic) as well as etiology, either congenital or acquired, later subdivided into traumatic, post-surgical, spontaneous, or pathological [4].

The management of pulmonary hernias depends on the hernia's underlying cause, size, location, and the patient's overall health. In some cases, nonsurgical management may be appropriate, such as observation or medical management of the underlying condition. However, surgical intervention may be necessary if the hernia is causing permanent symptoms or is at risk of becoming complicated. Surgical management is also indicated to control extreme pain or if the herniation leads to infection [5]. Hemoptysis and respiratory failure are common signs that indicate the need for urgent surgical intervention. Although possible, incarceration and infarction are uncommon, given the lung's elasticity. If the patient is asymptomatic, management is somewhat controversial, with the traditional method being observation but more recent trends favoring a more aggressive approach to prevent complications [4].

Conclusion

In conclusion, we have presented an interesting case and imaging of a patient with an iatrogenic lung hernia secondary to spinal fusion via thoracotomy. Although rare, it is a potentially severe complication of thoracic surgery. Early recognition and prompt assessment for treatment are essential to prevent severe complications and to ensure a successful outcome. Imaging studies, such as CT scans and MRI, play a critical role in diagnosing iatrogenic lung hernia, and surgical repair is the primary mode of treatment. Future studies are needed better to understand this condition's risk factors and incidence and improve diagnostic and therapeutic approaches.

Background

Fetoscopic endoluminal tracheal occlusion (FETO) improves the survival rate in fetuses with severe congenital diaphragmatic hernia (CDH). We hypothesize that prenatal therapies into the trachea during FETO can further improve outcomes. Here, we present an *ex vivo* microinjection technique with rat lung explants to study prenatal therapy with nanoparticles.

Methods

We used microsurgery to isolate lungs from rats on embryonic day 18. We injected chitosan nanoparticles loaded with fluorescein (FITC) into the trachea of the lung explants. We compared the difference in biodistribution of two types of nanoparticles, functionalized IgG-conjugated nanoparticles (IgG-nanoparticles) and bare nanoparticles after 24 h culture with immunofluorescence (IF). We used IF to mark lung epithelial cells with E-cadherin and to investigate an apoptosis (Active-caspase 3) and inflammatory marker (Interleukin, IL-6) and compared its abundance between the two experimental groups and control lung explants.

Results

We detected the presence of nanoparticles in the lung explants, and the relative number of nanoparticles to cells was 2.49 fold higher in IgG-nanoparticles than bare nanoparticles ($p < 0.001$). Active caspase-3 protein abundance was similar in the control, bare nanoparticles (1.20 fold higher), and IgG-nanoparticles (1.34 fold higher) groups ($p = 0.34$). Similarly, IL-6 protein abundance was not different in the control, bare nanoparticles (1.13 fold higher), and IgG-nanoparticles (1.12 fold higher) groups ($p = 0.33$).

Conclusions

Functionalized nanoparticles had a higher presence in lung cells and this did not result in more apoptosis or inflammation. Our proof-of-principle study will guide future research with therapies to improve lung development prenatally.

Introduction

Congenital diaphragmatic hernia (CDH) is associated with abnormal lung development resulting in lung hypoplasia and persistent pulmonary hypertension [1,2], and the incidence of CDH is 1 in 2000–5000 live births [[3], [4], [5]]. Recently, fetoscopic endoluminal tracheal occlusion (FETO) was shown to improve fetal lung growth and improve the survival rate in fetuses with severe congenital diaphragmatic hernia [6,7]. However, FETO has negative side effects for the mother and fetus and the mortality of CDH remains high.

To further improve outcomes and reduce the negative side effects of a surgical intervention, alternative prenatal medical therapies are desired. We previously demonstrated the effectiveness of microRNA-200b as a potential prenatal therapy for CDH [8]. However, it is evident carriers for a safe and site-directed delivery of microRNA therapies are required. Nanoparticles provide site-directed drug delivery maintaining good biocompatibility and protective effects of its payloads [9], and we have recently demonstrated that IgG-conjugated chitosan nanoparticles can cross placental epithelial cells in a transwell model have the potential to serve as a medical therapy to treat congenital diseases prenatally [10]. Moreover, nanoparticles enhance the stability of therapeutic agents and targeted delivery reduce their adverse effects, including those of miRNA and drugs [11,12].

Some animal models have demonstrated that nanoparticles can be a good option for drug delivery to a fetus [11,13,14], and Ullrich et al. reported a prenatal therapy with miRNA loaded nanoparticles in rat lungs [11]. To optimize the delivery of nanoparticles to the lungs of CDH babies, we hypothesized that injection techniques into the trachea during FETO have potential as a medical therapy. Intratracheal injections in adult animals has been reported [15,16], but the intratracheal injection of nanoparticles in fetal lungs and their biodistribution in animal models has not been investigated. Chitosan nanoparticles can achieve specific drug delivery to target tissues through surface modification with ligands like aptamers, peptides, and antibodies. We considered the prospect of utilizing this technology for prenatal therapy, particularly in the context of congenital diaphragmatic hernia (CDH) - to enable specific drug delivery to the fetal lung.

Here, we present an *ex vivo* microinjection technique using a FETO model of rat lung explants to study nanoparticles as a vehicle for prenatal drug delivery as an additive therapy to FETO.

Discussion

FETO is now being promoted for prenatal therapy of abnormal lung development in CDH, but FETO has negative side effects and the survival rate after FETO remains low. To further improve outcomes, alternative prenatal (medical) therapies are needed, and the use of nanoparticles for drug delivery to a fetus presents a promising option. In this study, we performed intratracheal administration of nanoparticles in rat lung explants with a microinjector, and confirmed the uptake of nanoparticles in

Conclusions

Our novel microinjection model to deliver nanoparticles effectively into the trachea with rat lung explants holds promising potential as a tool to study prenatal treatment for CDH, and provides insights into a new prenatal therapy. This model offers a controlled platform to investigate the potential of prenatal therapy with nanoparticles.

Case Description

A 61-year-old morbidly obese woman (weight: 91 kg, body mass index: 41) with a history of arterial hypertension and chronic obstructive pulmonary disease (COPD) and with a known preexisting para-

esophageal hernia was referred to our attention for management of early-stage lung adenocarcinoma of the left lower lobe (T1 cN0 M0) (Figure 1, *A*). No other lesions were found on whole body positron emission tomography scan. The patient complained only of occasional gastroesophageal reflux; for that her local gastroenterologist did not recommend surgical correction. After preoperative multidisciplinary assessment, the patient was scheduled for concomitant laparoscopic hernia repair and thoracoscopic lobectomy.

1. Download: Download high-res image (285KB)
2. Download: Download full-size image

Figure 1. A, Preoperative computed tomography scan showing the preexisting para-esophageal hernia and the lung cancer of *left* lower lobe. B, Postoperative computed tomography scan showing complete expansion of *left* upper lobe and no recurrence of para-esophageal hernia. ∗Lung cancer; ∗∗Paraesophageal hernia.

The patient was intubated with a double-lumen tube and the para-esophageal hernia was repaired through a standard laparoscopic approach. The hernia containing the stomach was reduced, and the hernia sac was excised. The gastric lipoma and gastric vessels were resected using LigaSure (Medtronic) up to the left crura of the diaphragm. The hiatal hernia defect was closed with interrupted sutures, followed by a Dor fundoplication. Then, the patient was placed in a right lateral decubitus position and a standard triportal thoracoscopy with anterior access was performed. The pulmonary ligament was carefully resected from the hernia sac using a Harmonic device (Ethicon Endo-Surgery Inc), and the inferior pulmonary vein was isolated and mechanically resected. Then, the fissure between the S5 and S7 segment, the A6 artery branch, the basal pyramid artery, and the lower bronchus were sequentially stapled. After retrieval of the specimen, a radical lymphadenectomy completed the procedure, and 1 chest tube for drainage was left in the pleural cavity. Video 1 summarizes the procedure.

Total operative duration was 235 minutes, with 100 minutes for hernia repair and 135 minutes for lobectomy. The total estimated blood loss was 270 mL and no intraoperative issues were found. Surgical pathology confirmed a pT1 cN0 M0 adenocarcinoma, with negative surgical margins. Postoperative course was unremarkable. Chest drainage was removed 3 days later, and the patient was discharged 5 days after the operation. At 1-month follow-up, a computed tomography scan (Figure 1, *B*) and barium swallow radiograph showed expansion of the remaining lobe without hernia recurrence. The patient did not have symptoms of reflux and was able to tolerate an oral diet. The patient was followed with computed tomography scan every 3 months for the first 2 years for lung cancer and with yearly esophagogram for hernia repair. The patient gave a written informed consent for this publication.

## Discussion

Thoracoscopic lobectomy is the recommended approach for management of early-stage lung cancer in high-risk patients at present, due to incurring less surgical trauma compared with thoracotomy,[1] but a preexisting para-esophageal hernia may increase the risk of postoperative morbidity and mortality. Ten previous cases reported intrathoracic gastric volvulus resulting from pneumonectomy or lobectomy associated with a known preexisting hiatal hernia.[2,3] Thus, we planned simultaneous surgical repair of the hernia and lobectomy. This approach possibly prevented complications from the presence of hernia after lobectomy while the concomitant lobectomy did not

increase the risk of postoperative complications (ie, esophageal or stomach perforation or acute dysphagia) related to the hernia repair itself.[4] Potential additional advantages were that ipsilateral pleural or pericardial injuries related to the hernia repair were managed during lobectomy concurrently and that 1 dose of anesthesia, and 1 course of postanesthesia recovery were used for performing 2 different operations that would have required 2 staged procedures, with potential delay of lung cancer treatment.

Toloza and colleagues[3] reported the feasibility and safety of contralateral combined hiatal hernia repair with lobectomy for lung cancer, whereas in our case, for the first time, this procedure was performed on the same site. Technically, the presence of ipsilateral lung cancer did not interfere with the hernia repair, whereas the reposition of the stomach into the abdominal cavity facilitated thoracoscopic lobectomy (Figure 2) because the hernia, protruding into the posterior mediastinum, could complicate the identification and resection of the lower pulmonary vein, especially in a patient with obesity, as was seen here.

1. Download: Download high-res image (295KB)
2. Download: Download full-size image

Figure 2. Intraoperative view showing the hernia sac, which adhered to the mediastinum and lower lobe, after hernia repair.

During hernia repair, great care was made to prevent iatrogenic communications between diaphragm and pleural cavity given the presence of ipsilateral lung cancer. For that, our strategy was not indicated in presence of ipsilateral pleural effusion, due to the potential risk of abdominal contamination from pleural effusion, in case of diaphragm injury. A Dor rather than a Toupet fundoplication was performed because it was associated with a reduction of operative time (due to less dissection).[5] It could be valuable because lobectomy should be performed after hernia repair.

Background

The severity of pulmonary hypoplasia is a main determinant of outcome for babies with congenital diaphragmatic hernia (CDH). Antenatal administration of extracellular vesicles derived from amniotic fluid stem cells (AFSC-EVs) has been shown to rescue morphological features of lung development in the rat nitrofen model of CDH. Herein, we evaluated whether AFSC-EV administration to fetal rats with CDH is associated with neonatal improvement in lung function.

Methods

AFSC-EVs were isolated by ultracentrifugation and characterized by size, morphology, and canonical marker expression. At embryonic (E) day 9.5, dams were gavaged with olive oil (control) or nitrofen to induce CDH. At E18.5, fetuses received an intra-amniotic injection of either saline or AFSC-EVs. At E21.5, rats were delivered and subjected to a tracheostomy for mechanical ventilation (flexiVent system). Groups were compared for lung compliance, resistance, Newtonian resistance, tissue damping and elastance. Lungs were evaluated for branching morphogenesis and collagen quantification.

Results

Compared to healthy control, saline-treated pups with CDH had fewer airspaces, more collagen deposition, and functionally exhibited reduced compliance and increased airway resistance, elastance, and tissue damping. Conversely, AFSC-EV administration resulted in improvement of lung mechanics (compliance, resistance, tissue damping, elastance) as well as lung branching morphogenesis and collagen deposition.

Conclusions

Our studies show that the rat nitrofen model reproduces lung function impairment similar to that of human babies with CDH. Antenatal administration of AFSC-EVs improves lung morphology and function in neonatal rats with CDH.

Introduction

The severity of arrested airway branching morphogenesis, impaired lung cell differentiation, and pulmonary vascular remodeling are primary determinants for the cardiorespiratory challenges that babies with congenital diaphragmatic hernia (CDH) face when transitioning from fetal to neonatal life [[1], [2], [3]]. These alterations result in compromised development of the alveolar-capillary interface, thus predisposing babies with CDH to respiratory distress syndrome and pulmonary hypertension [1,4,5]. The changes observed in CDH hypoplastic lungs directly affect tissue forces, resulting in poor lung compliance, increased airway resistance, and need for immediate endotracheal intubation for mechanical ventilation to establish adequate oxygenation in the newborn [1,2,6,7,12]. A major contributor for the progression of airway pathogenesis that directly affects mechanical airway properties is the abnormal deposition of collagen [8,9]. In CDH, the elevated collagen deposition has been associated with increased stiffness of lung parenchyma and decreased lung compliance [10,11]. Despite advances in neonatal care with implementation of ventilation protocols and randomized studies on optimal ventilation modes [[13], [14], [15]], it is well recognized that the postnatal management of infants with CDH remains a clinical challenge and improving lung development after birth is too late [16].

For this reason, several groups have been testing antenatal strategies to accelerate lung development *in utero* [16,17]. Promising results have been obtained with a regenerative medicine approach using amniotic fluid stem cells and their derivatives, including extracellular vesicles (AFSC-EVs) [[18], [19], [20], [21], [22], [23], [24]]. EVs are lipid-bound nanoparticles that carry small RNAs, proteins, and lipids and are considered the effectors of stem-cell paracrine signaling [25]. In rodent and human models of CDH, antenatal administration of rat and human AFSC derived extracellular vesicles (AFSC-EVs) rescues dysregulated signaling pathways relevant to lung development, promotes branching morphogenesis, improves epithelial cell and fibroblast differentiation, and reverses vascular remodeling with upregulation of angiogenic factors [23,24,[26], [27], [28]]. The effects of AFSC-EVs on growth, maturation, and vascularization of rat fetal hypoplastic lungs are mainly mediated by their RNA cargo, as enzymatically digesting AFSC-EV RNA or blocking miRNAs involved in lung development using antagomirs ablated their beneficial effects in multiple models [23,26]. AFSC-EV beneficial effects on lung development were observed at canalicular and saccular stages, timepoints amenable for human translation during the second and third trimester of gestation [27].

Overall, the results obtained thus far demonstrated the efficacy of AFSC-EV therapy to fetal hypoplastic lungs at a morphological, cellular, and molecular level. Towards clinical translation of this promising therapy, we aimed to evaluate whether the beneficial effects of AFSC-EV administration on fetal lung development were associated with an improvement in lung function in neonatal rats with CDH.

Section snippets

AFSC-EV isolation and characterization

c-kit$^{+}$ AFSCs were isolated from multiple Sprague-Dawley dams during mid-gestation, as previously reported [23,29,30]. AFSCs cultured in alpha-minimal Essential Media (αMEM) supplemented with 20% Chang supplements and 15% fetal bovine serum (FBS). At 80% confluency, media was changed to αMEM media plus 7.5% exosome-depleted FBS. After 18 h of culture medium was collected and subjected to differential centrifugation at 300 g and 1200 g to remove dead cells and debris. Cleared medium was passed

Results

At E21.5, we found no fetal demise in the group of control rats injected with saline. The mortality in nitrofen-exposed fetuses injected with saline (28%) was higher than those injected with AFSC-EVs (18%; Fig. 2B). CDH + saline pups had reduced body weight (n = 6; 4.4 ± 0.4 g) in comparison to control + saline pups (n = 9; 5.1 ± 0.5 g; $p < 0.03$; Fig. 2C). No difference in body weight was observed between CDH + saline and CDH + AFSC-EV pups (n = 6; 4.5 ± 0.4 g; $p = 0.819$; Fig. 2C).

Discussion

This study demonstrates that newborn rats with CDH have impaired lung function that can be improved by antenatal administration of AFSC-EVs. This is likely due to an improvement in branching morphogenesis, as well as a rescue of collagen levels in the lung.

The model used in our experiments is considered to be the most robust to reproduce features of human fetal pulmonary hypoplasia secondary to CDH [[34], [35], [36], [37]]. However, it is a non-surviving model and pups die within minutes after