

Case Report

Report of a case of bochdalek hernia in an adult presenting as chronic cough: review of literature and discussion

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ABSTRACT

Adult symptomatic bochdalek hernias (BH) are rare and account for 0.16%-6% of all diaphragmatic hernias. Chronic abdominal pain is commonest type of presentation. We have a patient of BH presented with chronic cough and breathlessness on exertion. Aim was to study clinical course and outcome of patient presented to our institute. We also would like to review relevant literature available online and compare it with our findings. 30-year-male from peripheral area had recurrent episodes of cough and breathlessness on exertion since 7 years. He was treated as a case of lower respiratory tract infection. We did chest radiogram which revealed presence of haziness in left lower zone with obliteration left costophrenic angle. His computerized tomography scan revealed defect of approximate size 43×45 mm in posterolateral aspect of left hemidiaphragm with omental herniation. Patient was operated by lap converted to open primary bochdalek hernia repair. His postoperative course was uneventful. Review of literature done revealed cough as a rare type of presentation. Laparoscopy is becoming emerging modality of treatment to do surgical correction. Adult onset diaphragmatic hernias have variable clinical manifestations. High index of clinical suspicion with imaging studies like contrast enhanced computed tomography (intravenous plus oral contrast) of chest and abdomen are required for timely diagnosis and treatment.

Keywords: Bochdalek hernia, Adult onset diaphragmatic hernia, Congenital diaphragmatic hernia, Laparoscopic diaphragmatic hernia repair, Open diaphragmatic hernia repair

INTRODUCTION

Presentation of a Bochdalek hernia (BH) in an adult is exceptionally rare. Symptomatic adult BH account for 0.17%-6% of all diaphragmatic hernias.¹ Unusually, in adults, BH get detected as an incidental finding on contrast enhanced computed tomography (CECT) scan. Alternatively, it may be diagnosed only after complications occur. The clinical presentation of adult BH is varied and is mainly confined to the respiratory or gastrointestinal systems. We are presenting a report of a case of adult BH presenting as chronic cough along with a brief review of clinical literature, diagnostic and therapeutic options for adult diaphragmatic hernias.

CASE REPORT

30-year male patient presented to us with recurrent episodes of cough and breathlessness on exertion. Symptoms started almost 7 years ago. There was no history of trauma, tuberculosis or any respiratory problem. As there were no other associated symptoms, he was treated as a case of lower respiratory tract infections several times. Recently his symptoms increased and he came to us for further evaluation and treatment.

On clinical examination his per abdominal examination was normal. On auscultation, air entry was reduced in left lower zones. As shown in Figure 1, his X-ray chest revealed left lower zone haziness and blunting of

costophrenic angle. Ultrasonography of abdomen showed hyper-echoic lesion in left lower zone of chest. His contrast enhanced computed tomography (CECT) (intravenous plus oral contrast) of chest and abdomen (Figures 2 and 3 respectively) revealed 3.6 cm a defect in left posterolateral hemidiaphragm through which omentum measuring 4.4×11.7 cm seen herniating into left hemithorax with passive atelectasis of underlying left lung parenchyma.



Figure 1: Initial X-ray chest showing left lower zone haziness and blunting of costophrenic angle.

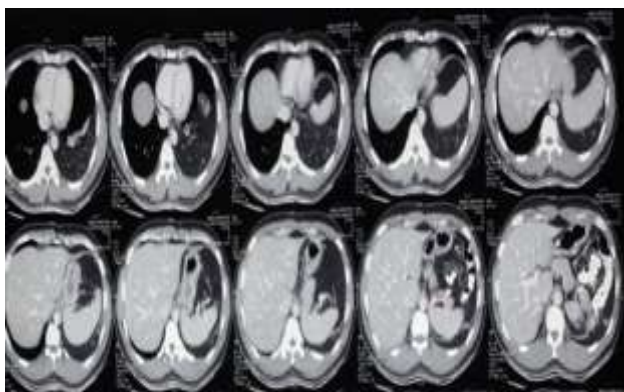


Figure 2: Contrast enhanced computed tomography chest showing omental herniation into left hemithorax with passive atelectasis of underlying left lung parenchyma.

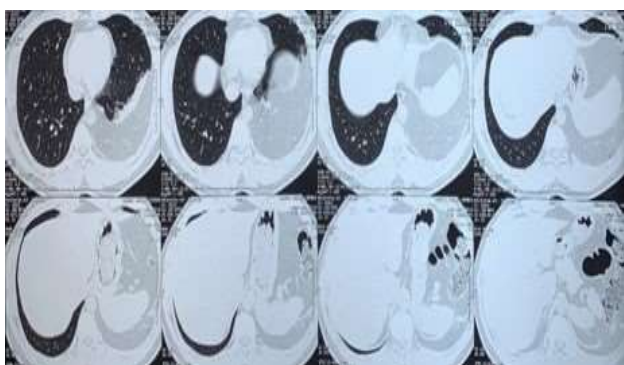


Figure 3: Contrast enhanced computed tomography of abdomen defect in left posterolateral hemidiaphragm with omental herniation.

Patient was planned for laparoscopic reduction with primary closure of the defect. Intraoperatively defect of 4×4 cm was present lateral to the spleen. Omentum was migrated with adhesions to diaphragm and surrounding structures (Figure 4). Splenic flexure was pulled towards the defect. Laparoscopic adhesiolysis with reduction of contents was carried out. Thoracic examination was done to see the thoracic cavity and the lung, it was found to be normal. As the defect was extreme posterolateral in position proper closure of defect with laparoscopy was technically difficult; subcostal incision was taken and repair of defect was done with multiple interrupted sutures with no-1 loop ethilon (Figures 5 and 6).



Figure 4: Intraoperative image showing omental migration.



Figure 5: Intraoperative image showing defect in diaphragm after reduction of content.



Figure 6: Intraoperative image showing closure of defect.

Intercostal drainage tube was placed. Postoperative course was uneventful and his symptoms showed improvement. His X ray chest taken during postoperative period showed disappearance of haziness in left lower zone with expansion of lung (Figure 7).



Figure 7: Post-operative X ray showing complete reduction of content with expansion of thoracic cavity.

DISCUSSION

Diaphragm is a muscular sheet separating the thorax from the abdomen and is composed of two domains: costal and crural diaphragm (Figure 8). The costal diaphragm is a thin domed sheet of muscle composed of a radial array of myofibers extending laterally from the ribs and medially to a central tendon. The crural diaphragm is thicker and located more posteriorly (dorsally), where it attaches to the vertebrae and surrounds the oesophagus and aorta. Medially, the myofibers of both the costal and crural muscles insert into the central tendon. The central tendon is located at the apex of the domed diaphragm, holding the diaphragm muscle domains together. Caudally, it attaches to the liver via the falciform and coronary ligaments.

Embryologically, the diaphragm develops from multiple embryonic sources. The muscle and its associated connective tissue and central tendon develop from three sources: the septum transversum, the pleuroperitoneal folds, and the somites. The septum transversum is the first structure present in the developing diaphragm and serves as the initial barrier between the thoracic and abdominal cavities in all vertebrates. The septum transversum is a thin, mesodermal sheet of tissue that separates the heart from the liver. The pleuroperitoneal folds (also referred to as the post-hepatic mesenchymal plate) are the second important component of the developing diaphragm. They are two transients, pyramidal-shaped structures lying on either side of the oesophagus that protrude from the body wall between the pleural and peritoneal cavities. These pyramidal structures are called as pleuroperitoneal canals. These canals close over a period of time. Similar to trunk and limb muscles, the somites are the source of the diaphragm's muscle cells.

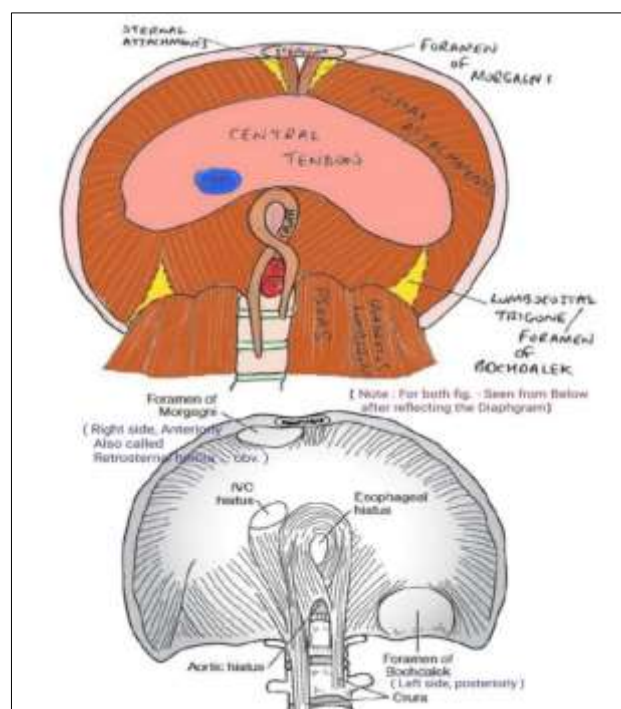


Figure 8: Diagrammatic image of diaphragm.

Kluth et al studied embryology and formation of congenital diaphragmatic hernias (CDH) in detail.² Accordingly, CDH are a group of diaphragmatic malformations due to a failure of the pleuroperitoneal canals to close at the end of the embryonic period (8th gestational week). This stops the process of fusion of the septum transversum with pleuroperitoneal folds of the diaphragm keeping a weak area in the diaphragm from which herniation happens. CDH was first described by Lazare Riviere in a postmortem examination of a 24-year-old male. In 1754, George Macaulay reported the first neonate with CDH, also in a necropsy finding in an infant who died from respiratory failure.³

The most common type of CDH is a Bochdalek hernia; other types include Morgagni hernia, diaphragm eventration and central tendon defects of the diaphragm. Depending on the location of the defect in the diaphragm, the hernias can be classified into different types. Bochdalek hernias result from a defect in the posterolateral part of the diaphragm and are the most common type (70% to 75%), with the majority occurring on the left side and less frequently on the right side. Morgagni hernias result from a defect in the anteromedial part of the diaphragm (20% to 25%), and central hernias account for 2% to 5%.⁴

Malformation of the diaphragm allows the abdominal organs to push into the chest cavity, hindering proper lung formation. The intra-thoracic negative intrapleural pressure and the intra-abdominal positive pressure play a main role in migration of viscera into the thoracic cavity.

Symptoms of diaphragmatic hernias varies with the age at which they are presenting.

Neonates with CDH usually present early in the first few hours of life with respiratory distress due to overcrowding of the thoracic cavity. The respiratory distress accompanied by the CDH may be mild. Occasionally the accompanied respiratory distress is so severe it can be life-threatening. Adult onset diaphragmatic hernia is a rare condition with variable clinical manifestations. The most frequent symptom in adults is mild discomfort and patients are mainly asymptomatic making diagnosis difficult.⁵ Since the clinical presentation is varied, a high degree of clinical suspicion coupled with imaging studies are required for timely diagnosis and treatment.

The diagnosis of the disorder can be established using radiological techniques. Chest radiograph is usually the first imaging procedure but it is of limited value on depicting the diaphragmatic defect. The hernia may appear either as a soft-tissue opacity at the lung base or as a solitary, smooth, round lesion in the posterior costophrenic recess. Contrast studies like barium meal follow through may confirm the diagnosis in some cases. Abdominal ultrasound sometimes may depict the disruption of diaphragmatic continuity with the associated herniated organs, distinguishing fatty tissue from liver and loops of bowel.⁶ CT is the procedure of choice for demonstrating a bochdalek hernia. With routine CT scan, Killeen et al demonstrated sensitivities of 50% for right-sided and 78% for left-sided bochdalek hernia detection.⁷

Surgical reduction of the organs into abdominal cavity and closure of the hernial defects with or without mesh is mainstay of treatment. Abdominal, thoracic and both approaches were possible depending on the experience of the surgeon. Recently, thoracoscopic and/or laparoscopic surgery for treatment of adult BH is becoming more popular due to small scar, less pain and early recovery. However, there is no study comparing open and laparoscopic surgery. Several methods can be used to repair the defects. Some authors have reported the use of prosthetic material as reinforcement, whereas other authors preferred simple suturing of the defect. It is generally agreed that defects larger than 20–30 cm² require a prosthesis. However, no study has reported an improved method of repairing diaphragmatic defects.⁸ Although the outcome of diaphragmatic hernias has not been studied properly in one of the reported study the complication rate of hernia repair was 19%, and mortality rate was 4.5%.⁹

In our case, patient had symptom of cough for seven years which was treated as a case of recurrent lower respiratory tract infection. Patient reported to us on aggravation of symptoms. CECT (intravenous plus oral contrast) of chest and abdomen was diagnostic which revealed the presence of diaphragmatic defect with herniation of omentum into the thoracic cavity. In this case, high index of suspicion would have helped previous physicians to diagnose BH at early stage.

We completed laparoscopic reduction of contents but due to the extreme posterolateral position of the defect there was difficulty in laparoscopic closure of the defect. Decision to convert to an open procedure was taken to avoid inadequate closure of defect leading to possibility of recurrence in future. A left subcostal incision was taken and primary closure done with interrupted sutures with no-1 loop ethilon. His post-operative recovery was uneventful and at 3 months follow up, the patient is stable with no recurrence of symptoms.

CONCLUSION

Adult onset diaphragmatic hernias have variable clinical manifestations. High index of clinical suspicion with imaging studies like CECT (intravenous plus oral contrast) of chest and abdomen are required for timely diagnosis and treatment.

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