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Case Report

A rare case of late-presenting congenital diaphragmatic hernia in an adult patient

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ABSTRACT

Congenital diaphragmatic hernia (CDH) refers to a true breach in the continuity of the diaphragm, allowing herniation of abdominal contents into the thoracic cavity. Usually, it presents in the neonatal stage. Adult presentations are rare. Most adult cases of diaphragmatic hernia are of traumatic etiology. Most adult CDH cases are asymptomatic and detected incidentally on imaging. Symptomatic adult CDH cases are uncommon. Bochdalek hernias are more common than the Morgagni type. Most of them are left-sided due to embryonic factors. We present an uncommon case of symptomatic, non-traumatic, left-sided CDH in an adult patient. A 20-year-old male presented with left hypochondriac pain for eight days and left-sided chest pain for three days in the absence of any trauma or comorbidities. On examination, the left chest showed reduced chest movement, diminished breath sounds, and gurgling on auscultation. Chest X-ray showed air-fluid levels in the left hemithorax and right mediastinal shift. Contrast-enhanced computed tomography showed an overdistended stomach in the left hemithorax. Upper midline laparotomy revealed an 8×7 cm defect in the posterior left hemidiaphragm, consistent with a Bochdalek hernia. The stomach and spleen were herniated into the left hemithorax. They were reduced into the peritoneal cavity, and the defect was repaired with primary closure. Diaphragmatic plication was done to reinforce the thinned left hemidiaphragm. This case underscores the importance of considering CDH in adults presenting with thoracoabdominal symptoms, especially in the absence of any trauma. Surgical exploration provides the most definitive diagnosis in cases of equivocal imaging in adult CDH patients.

Keywords: Congenital diaphragmatic hernia, Bochdalek hernia, Adult patient, Non-traumatic

INTRODUCTION

Diaphragmatic hernia refers to a true breach in the continuity of the diaphragm, allowing the herniation of abdominal contents into the thoracic cavity. The incidence of non-hiatal diaphragmatic hernias is 2.3 per 10,000 live births.[1] Congenital diaphragmatic hernias (CDH) are most commonly diagnosed in the paediatric population. The diagnosis in the adult population is rare with an incidence rate of 0.17%.[2] In adults, non-traumatic diaphragmatic hernias are less prevalent than those due to traumatic etiology,[3] with Bochdalek hernias being more prevalent than Morgagni type.[4] Often the defect is seen on the left side due to the underlying embryogenesis mechanism.

Most of the time, these are incidental findings; very rarely do patients present with gastrointestinal or respiratory symptoms.[3] Computed tomography is the cornerstone of radiological diagnosis of CDH.[5] We present the case of a symptomatic, non-traumatic left-sided congenital

diaphragmatic hernia in a 20-year-old adult patient followed by a discussion on this uncommon entity.

CASE REPORT

A 20-year-old male patient presented to our hospital with abdominal pain for eight days and left-sided chest pain for three days. The patient was relatively asymptomatic before eight days after which he developed pain in the left hypochondriac region which was colicky, intermittent, and non-radiating.

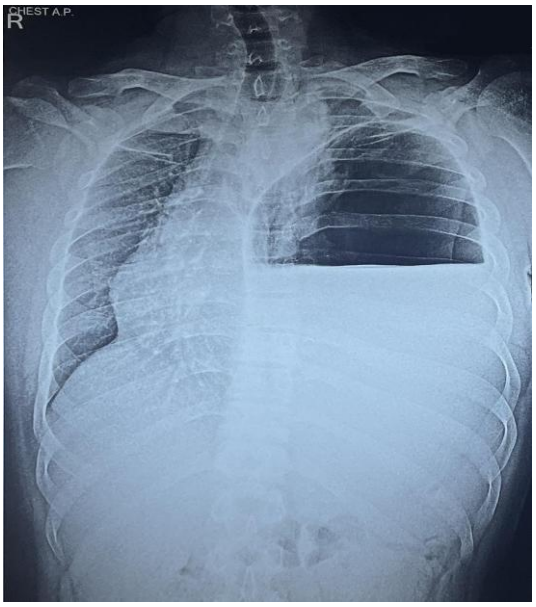

Figure 1: Chest X-ray showing air-fluid levels in left-sided chest and right mediastinal shift.

After five days, the patient developed chest pain which was dull aching and non-radiating. There was no associated fever, vomiting, jaundice, weight loss, voiding difficulty, diarrhoea, history of trauma, melana, per rectal bleeding, or cough for a long time.

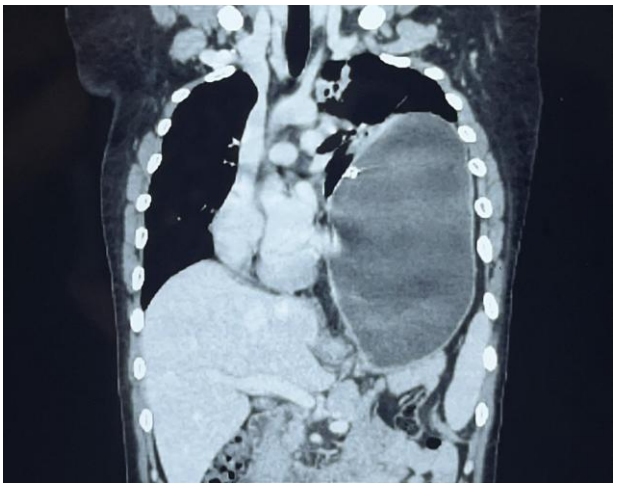

Figure 2: Contrast enhanced CT showing overdistended stomach in left hemithorax.

The patient had no other co-morbidities, medical or surgical history. The patient had been a chronic smoker for 3 years, consuming approximately 5-6 cigarettes a day. The patient was averagely built and had a body mass index (BMI) of 28.7 kg/m^2. On examination, chest movement and tactile vocal fremitus were reduced on the left side. Apex beat was not palpated in the left 5th intercostal space. On auscultation, the left middle and left lower chest regions revealed a gurgling sound and the left upper region revealed diminished breath sounds. Right-sided auscultatory findings were normal.

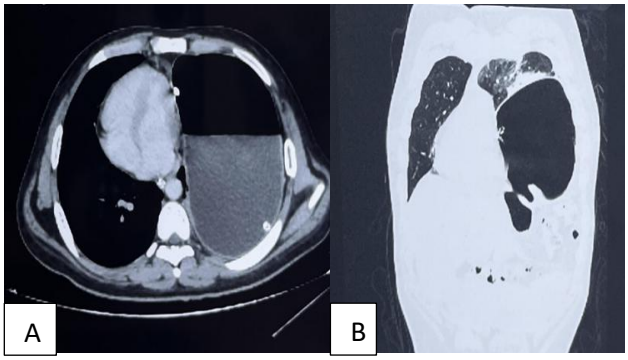

Figure 3: (A) CECT showing deviation of heart to right side, suggesting right mediastinal shift, (B) areas of increased lung attenuation seen in left upper lobe.

Chest X-ray revealed air fluid levels in the left-sided chest. Heart, trachea, and mediastinum shifted to the right, suggesting right mediastinal shift (Figure 1). Ultrasound revealed over distended stomach. Contrast-enhanced computed tomography (CECT) demonstrated an overdistended stomach present in the left hemithorax (Figure 2). It was also suggestive of a mesentero-axial type of gastric volvulus. Mediastinal shift towards the right side was noted (Figure 3A) with passive collapse of the left lower lobe and lingular segment of the left upper lobe. A few atelectatic bands and areas of increased attenuation were noted in the remnant of the left upper lobe (Figure 3B). A nasogastric tube was inserted for decompression of the overdistended stomach.

An upper midline laparotomy was performed. Intraoperatively approximately 8x7 cm defect was noted over the posterior surface of the left hemidiaphragm (Figure 4A) through which the stomach and spleen were found to be herniating into the left thoracic cavity. Hence, intraoperatively a diagnosis of left Bochdalek hernia was likely. No gastric volvulus was found intraoperatively. The stomach and spleen were reduced into the peritoneal cavity after freeing them from all attachments. Primary closure of the diaphragmatic defect was done using Prolene 2-0. The remaining left hemidiaphragm was thinned out for which diaphragmatic plication was done using Prolene 2-0 (Figure 4B). The integrity of the left pleural cavity was confirmed. A 32FG Romo ADK drain was inserted in the splenic fossa and brought out from the left side and fixed to the skin using silk No.1. Sheath closed using polydiaxone (PDS) No.1 in a continuous non-locking

manner with intermittent Aberdeen knots. Skin closed with vertical mattress sutures using epimide 2-0. A sterile dressing was applied. The postoperative period was uneventful and the patient was discharged on postoperative day 4.

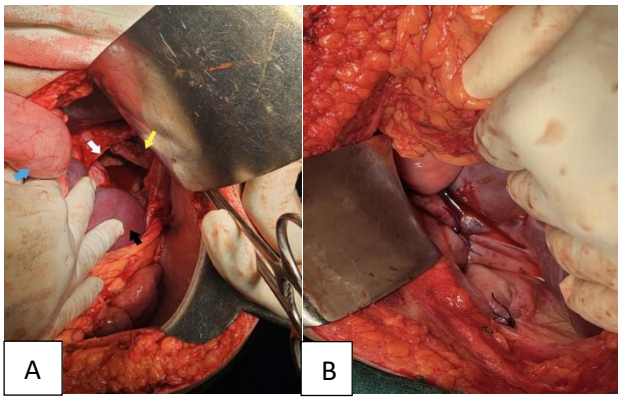

Figure 4 (A): 8x7 cm hernial defect seen over the posterior surface of the left hemidiaphragm (white arrow) with the lungs (yellow arrow) visible through the defect. Stomach (blue arrow) and spleen (black arrow) have been reduced into the peritoneal cavity, (B) diaphragmatic plication done to reinforce the thinned left hemidiaphragm after the defect was repaired.

DISCUSSION

Congenital diaphragmatic hernias are of two types, Bochdalek (posterolateral) and Morgagni (anteromedial) with Bochdalek being the more prevalent type.[6] Pathogenesis involves the failure of the pleuroperitoneal membrane to contribute to the diaphragm. Earlier closure of the right-sided membrane and the presence of hepatic buffer make right-sided CDH less common than left.[3] It is proposed that small congenital defects may widen overtime due to compounding intra-abdominal and intra-thoracic pressure over the years, causing eventual gross visceral herniation, and finally become symptomatic in adulthood, as in our case.[7]Herniation of abdominal viscera, such as stomach, liver, bowel, spleen, or omentum is commonly seen.[8,9] This can precipitate chest or abdominal pain and pressure, dyspnoea, dysphagia, indigestion, visceral obstruction, strangulation, hypertension, and GERD.[10]

Hence, timely intervention is imperative. Due to close symptomatology, it is vital to distinguish CDH from similarly presenting conditions such as diaphragmatic eventration, congenital cystic adenomatoid malformations, bronchogenic cyst, bronchial atresia, bronchopulmonary sequestration and teratoma via robust imaging.[2,11] In our case, the gross visualisation of an overdistended stomach in the left hemithorax nearly ruled out these differentials. Useful imaging modalities in CDH are chest X-ray, CT scan, MRI, and barium studies. X-ray findings usually show intrathoracic viscera with contralateral mediastinal shift. Contrast CT demonstrates diaphragmatic discontinuity. CT scan shows a 14-82 % sensitivity and 87% specificity in CDH cases.[7,12] Though imaging is a highly important investigation in CDH and related pathologies, it is difficult to establish an accurate diagnosis preoperatively, as in our case where the presence of a diaphragmatic defect was confirmed only intraoperatively and with not only the stomach, but also with the spleen herniating in the left hemithorax.[13] In our case, the hernial defect was not evident on CECT. The splenic herniation was also not reflected on CECT. The gastric volvulus suggested by CECT was not found intraoperatively in this patient. Sometimes, due to radiological similarities in appearance, a close differential pathology, such as diaphragmatic eventation, may be incorrectly reported as a diaphragmatic hernia. Though imaging is a highly important investigation in CDH and related pathologies, it is difficult to establish an accurate diagnosis preoperatively, as in our case where the presence of a diaphragmatic defect was confirmed only intraoperatively, and with not only the stomach, but also the spleen herniating in the left hemithorax.[11]

Surgical approaches include thoracotomy, laparotomy, and minimally invasive surgery via thoracoscopy or laparoscopy.[10] Small defects up to 10cm^2 are repaired by primary closure and larger defects using mesh hernioplasty.[14] In our case upper midline laparotomy was performed and primary closure of the defect was done. Also, a diaphragmatic plication surgery was performed to reinforce the thinned left hemidiaphragm. The postoperative complications such as pneumothorax, hydrothorax, and abdominal compartment syndrome can be seen after CDH repair.[6] Studies suggest that both transthoracic and transabdominal techniques yield similar results in terms of postoperative complications and recurrence rates.[15]

CONCLUSION

Non-traumatic symptomatic congenital diaphragmatic hernias in adult populations are rare. This case highlights that congenital diaphragmatic malformations cannot be completely ruled out in adult patients presenting with thoracic or abdominal symptoms, especially in the absence of obvious trauma. Small defects can increase in size due to compounded pressure over time and can present with symptoms in adulthood. The most accurate diagnosis of CDH in adults can be made only intraoperatively after surgical exploration.

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