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Bochdalek Diaphragmatic Hernia Rupture in Pregnancy: A Case Report

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Abstract

Objective: Dyspnea in pregnancy and peri-partum period is a relatively common symptom, potentially caused by a number of diseases, asthma, pulmonary infection, embolism and heart disease. Diaphragmatic hernia is a rare cause, more likely to be misdiagnosed, with a high maternal and fetal mortality risk; therefore, knowledge of this entity is very important.

Methods: We present the case of a nulliparous woman at 35+2 weeks gestation, with a history of asthma, self-referred to the emergency room with upper and lower abdominal pain radiating to the back, vomiting, tachycardia and fetal bradycardia. The ultimate diagnosis was a Bochdalek hernia rupture.

Results: The patient was misdiagnosed initially with suspected placental abruption and underwent an emergency caesarean section under general anesthesia, complicated by aspiration pneumonia. Further investigations revealed the rupture of a congenital diaphragmatic hernia.

Conclusion: The differential diagnosis for severe dyspnoea in pregnancy should include diaphragmatic hernia as a potential albeit rare cause.

Introduction

Congenital diaphragmatic hernia has an incidence of 1:2000-1:12,500 live births and accounts for 8% of all the major congenital anomalies [1]. Bochdalek Hernia (BH) is the commonest form of congenital diaphragmatic hernia and usually a perinatal pathology. However, in the literature, there are case reports of BH presenting in adults, with an incidence reported to be 0.17% [1]. Rupture of a diaphragmatic hernia is a very rare, but significant complication, with an increased maternal mortality rate (up to 10%) and fetal mortality rate (up to 13%). It normally occurs in the peripartum period, generally because of a delayed diagnosis and requires a multidisciplinary team approach involving obstetricians, cardiothoracic surgeons, anesthetists and radiologists [2].

Diaphragmatic hernia is classified as hiatal, congenital or traumatic, with the first being the most common type [3]. The diaphragm development starts in the 4th weeks of gestation when the central tendinous area forms from the septum transversum, which separates the thoracic and abdominal cavities and finishes by the 8th week with the fusion of the lumbar and costal muscle groups in the posterolateral diaphragm. As this is the final stage of the development of the diaphragm, it represents a vulnerable loci and it is at increased risk of a hernia formation. Complete closure occurs in the right side before the left and this is a fact that may contribute to the left sided Bochdalek hernias [4]. Another theory would be that the liver protects the right side of the diaphragm and this is why these hernias are more likely to occur on the left [5]. In a study by Mullins et al. a higher incidence of BH was found in women (77%) and pregnancy was found to be the most common precipitating factor in the rupture of a BH (34%) [6].

Adult patients presenting with BH may also have associated congenital anomalies (12%) [3]. These more common anomalies include malrotation and incomplete attachment of the caecum, hepatic hypoplasia, bifid liver, pulmonary hypoplasia. The approach to repair the BH depends on the presentation (emergency or elective), size and site of the defect and the presence of complications. In recent years, there has been an increase in the use of the laparoscopic surgical approach in the management of BH [1]. Surgical repair performed in an elective setting is the treatment of choice; the mortality in elective surgery is 5%, compared to 32% following emergency surgery.

Case Presentation

A 36 years old primiparous woman at 35+2 weeks gestation with a history of asthma using

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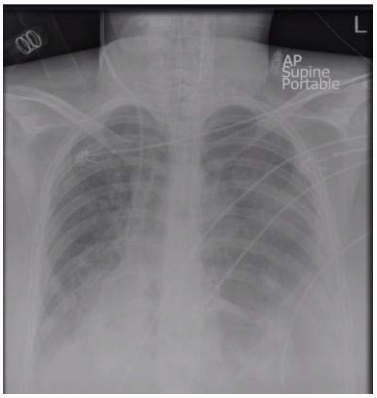

Figure 1: AP Portable Chest X-ray Endotracheal tube, right central venous access in situ. Day 1 post caesarean section. Mediastinal shift to the right side. Right lower lobe atelectasis/consolidation with patchy opacification involving both lungs likely due to aspiration.

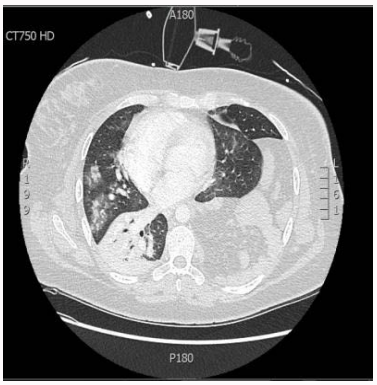

Figure 2: CT Thorax day 1 post caesarean section. Jejunal, ileal loops & mesentery in left hemithorax consistent with left diaphragmatic hernia and right mediastinal shift. Patchy ground glass opacification in both lungs with right lower lobe consolidation likely due to aspiration.

bronchodilators self referred to the emergency room with severe generalized abdominal pain radiating to the back with vomiting. Examination revealed a maternal heart rate of 130 beats/min and respiratory rate of 24 cycles/min. Her abdomen was soft with non-specific tenderness. Fetal bradycardia of 80 beats/min was noted on CTG. An emergency caesarean section for suspected placental abruption was performed under general anesthesia. Significant pulmonary aspiration occurred on induction of anesthesia and over 1 litre bilious fluid was aspirated through an oro-gastric tube. A live male infant was delivered with no evidence of placental abruption.

Postoperatively, she was managed in the intensive care unit for five days. A chest X-ray showed mediastinal shift to the right side with consolidation/atelectasis of the right lower lobe (Figure 1). Initial clinical deterioration led to increasing ventilation requirement. A subsequent CT thorax showed jejunal and ileal loops and mesentery displaced into the left hemithorax consistent with a large left-sided diaphragmatic hernia (Figure 2). This was most likely congenital in aetiology.

She was treated with broad-spectrum antibiotics for aspiration pneumonia while intubated and sedated. Ventilatory support was weaned over next 24 hours. Following extubation, high flow O_2 and intermittent BiPAP was commenced. Renal indices and electrolytes remained normal. She was discharged from ICU on high flow O_2.

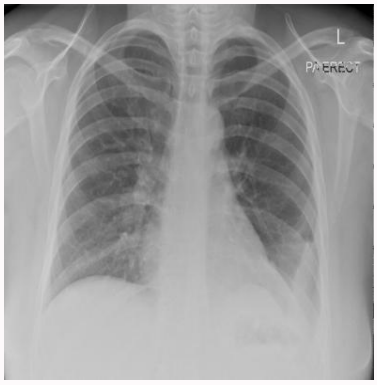

Figure 3: PA Chest X-ray day 7 post laparoscopic diaphragmatic hernia repair. The left lung is now almost fully inflated with small residual loculated hydropneumothorax in the left lower hemithorax.

Following review by the cardio-thoracic surgery and upper gastro-intestinal surgery teams the decision was made to treat the pulmonary aspiration first and the diaphragmatic hernia at a later date. On day 12 postpartum, she was discharged home.

Ten weeks postpartum, an elective laparoscopic left hemi-diaphragmatic hernia repair was performed. There was a significant 5x5 cm posterolateral left diaphragmatic defect consistent with Bochdalek hernia through which her entire small bowel, ascending colon and transverse colon had herniated into the left hemithorax.

The small bowel and colon were successfully reduced into the abdominal cavity. The defect was closed primarily and reinforced with a Gore-Tex mesh.

Day 2 post laparoscopy, the chest X-ray demonstrated suboptimal re-expansion of the left lung. A pigtail catheter was inserted and the chest drain was placed on high-pressure suction. A week following her laparoscopy, chest X-ray showed the left upper lobe had fully re-expanded but the left lower lobe was trapped and there was a persistent postero-lateral space which was air and fluid filled (Figure 3). Since the procedure her breathing has much improved, but the persistent hydro-pneumothorax places her at increased risk of pulmonary infections in the future.

Discussion

Dyspnoea in pregnancy and the peripartum period is a relatively common symptom, potentially caused by a number of diseases including respiratory tract infections, asthma, pulmonary embolism, pulmonary oedema and heart disease including peripartum cardiomyopathy. By contrast, BH is a far less common cause of peri-partum dyspnea, but its recognition is of utmost importance as it has an increased risk of both maternal and perinatal mortality.

Pregnancy is a significant predisposing factor and when BH is found incidentally in pregnant women, it may warrant an elective repair, preferably in the second trimester. Frequently, these cases present acutely with complications. CT and MRI are useful diagnostic tools.

We present a case of a ruptured undiagnosed diaphragmatic hernia in pregnancy. The presentation in this case suggested a placental abruption. The clinical situation was complicated by a pulmonary aspiration secondary to the general anesthesia, associated with a bowel obstruction. Further investigations identified the

rupture of a Bochdalek hernia and the presence of small bowel loops in the thoracic cavity. In view of the post-operative complications, the decision was made to stabilize the patient and treat the aspiration pneumonia, followed by an elective laparoscopic hernia repair.

The primary repair and mesh reinforcement performed via laparoscopy is in keeping with current practice with the associated well established benefits. Pneumothorax has been recognized as a potential complication following repair with a case incidence of 0.8% [1]. In view of the high risk of complications, a multidisciplinary team formed by obstetricians, radiologists, cardio-thoracic surgeons, upper gastro-intestinal surgeons and anesthetists is required in these cases.

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