Diaphragmatic Rupture with Background Diaphragmatic Eventration in Pregnancy

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ABSTRACT-

Introduction-Diaphragmatic rupture can result from trauma or, in rare cases, occur spontaneously. Diaphragmatic eventration is an uncommon condition characterized by partial or complete thinning of the diaphragm's musculature. This can arise from a congenital defect in the diaphragm or as a secondary consequence of phrenic nerve injury. Often asymptomatic, it is frequently underdiagnosed. A spontaneous rupture associated with congenital eventration is extremely rare. Diagnosis primarily relies on chest radiography or computed tomography scans of the chest.

Case presentation -A 32-year-old G3P1 woman at 8 weeks of gestation presented at the Emergency Department with abdominal pain, vomiting, and difficulty in breathing. Plain chest x-ray showed strong evidence of diaphragmatic eventration with abdominal contents in the left hemithorax. She subsequently had repair of diaphragmatic rupture with good outcome.

Discussion-Diaphragmatic eventration is a rare but significant condition in which abdominal organs shift into the chest cavity due to dysfunction or weakness of the diaphragm. This condition can rupture either spontaneously or following a traumatic injury. Prompt recognition is critical, especially when there is a risk of ischaemic or necrotic tissue, which can lead to serious complications. Swift diagnosis and effective treatment are vital to achieving the best patient outcomes, underscoring healthcare professionals' need to remain vigilant and informed about this serious condition.

Conclusion -A prompt response to this clinical scenario is essential for achieving optimal outcomes. This case clearly demonstrates the necessity of immediate recognition, intervention and effective management in pregnancy situations.

Keywords: Viscus Herniation, Diaphragmatic Rupture, Diaphragmatic Eventration, Plication

I. INTRODUCTION

Diaphragmatic rupture typically occurs as a result of congenital defects in the diaphragm or due to traumatic injuries to the abdomen or chest[.1]. Both gastric and diaphragmatic ruptures are quite uncommon, and their simultaneous occurrence is even rarer[1,2.] Diaphragmatic eventration is characterized by an abnormal elevation of the diaphragm while maintaining normal peripheral attachments. The condition can be either congenital or acquired. Acquired cases typically arise from trauma to the phrenic nerve, whether due to mechanical injury or surgical intervention. Additionally, it can result from compression caused by space-occupying lesions in the thorax, as well as multiple infections and inflammatory conditions that damage the phrenic nerve. The predominant cause of acquired eventration is injury to the phrenic nerve, which may result from either traumatic birth or thoracic surgery performed to address congenital heart disease. An injury to the phrenic nerve can cause the diaphragm to shift upward; over time, this can lead to the replacement of muscular tissue with stiff, fibroelastic tissue, resulting in diaphragmatic eventration[3,4]. In contrast, congenital eventration arises from either an underdeveloped diaphragmatic muscle or the absence of the phrenic nerve[3].

The estimated prevalence is 1 in 10,000 live births, with the condition typically occurring on the left side[4].

Unilateral eventration is linked to Beckwith-Wiedemann syndrome and trisomies 13, 14, 15, or 18. Bilateral eventration is associated with toxoplasmosis, cytomegalovirus, arthrogryposis, and Werdnig-Hoffman disease[5].

Gastric rupture can occur due to factors like excessive fluid intake, nasal cannula oxygen therapy,

or pyloric obstruction from tumors or stenosis, with a mortality rate of up to 85%[2].
Diaphragmatic and gastric ruptures during pregnancy are uncommon occurrences that predominantly manifest in the third trimester, particularly in individuals with a pre-existing diaphragmatic hernia. The associated mortality rate during pregnancy is significantly high, and there are very few documented cases in which both the mother and child survive these complications.[2,6]

Diaphragmatic eventration is rare and even rarer is non traumatic diaphragmatic rupture in first trimester.
Most adults with diaphragmatic eventration are asymptomatic, with diagnosis often made incidentally via chest radiography. For those who are symptomatic, the primary complaint is dyspnea, while over 50% of adults show no symptoms and 25% experience mild exertional dyspnea. Symptoms may include shortness of breath and gastrointestinal issues such as epigastric pain, burning sensation, regurgitation, nausea, belching, and abdominal fullness.

The management of diaphragmatic eventration is contingent upon the etiology and severity of the condition. In cases that are asymptomatic or exhibit mild symptoms, a regimen of supportive care is typically recommended. In instances where hypoxemia is present, it is essential to provide oxygen supplementation[7]. Conversely, urgent surgical intervention with primary closure is deemed the optimal approach for the management of diaphragmatic rupture[8].

II. CASE REPORT

A 32-year-old woman, G3P1, presented to the gynecological emergency department at 8 weeks of gestation with abdominal pain and frequent vomiting lasting for 5 days, accompanied by difficulty in breathing for the past 24 hours. She has no history of smoking and reports no trauma, accidents, or similar episodes of dyspnea in the past. Notably, there is a history of left hemidiaphragmatic elevation observed in a pre-employment chest X-ray performed two years prior to presentation.

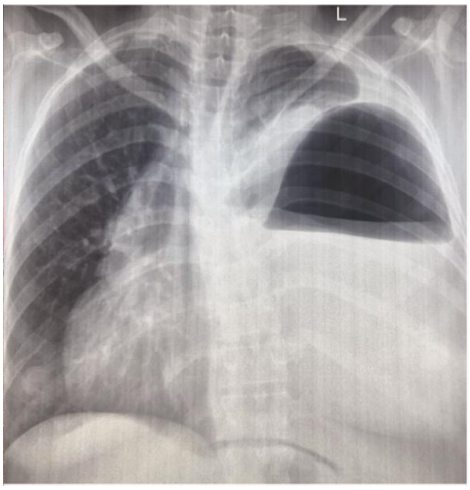
Upon examination, the patient presented with dyspnea and tachypnea, with a respiratory rate of 30 breaths per minute. There was reduced air entry in the left hemithorax. At the time of her presentation to the gynecological Emergency Department, she exhibited a blood pressure of 100/60 mmHg, a pulse of 102 per minute, and a temperature of 37.7°C. The abdominal examination revealed distension, generalized tenderness, and guarding Chest X-ray (Figure 1) showed an area of hyperlucency devoid of lung markings, with a crescentic dome just sparing the apex of the left lung. The gastric bubble with its fluid level was seen in the left hemithorax.

She was resuscitated with intravenous fluids, antibiotics, and oxygen therapy. Her case management involved a team of Gynaecological, Cardiothoracic, and General Surgeons. She received counseling regarding her condition, the necessity for surgical intervention, the impact of sepsis on the fetus, and the challenges that her pregnancy posed to her recovery.

She underwent manual vacuum aspiration of the fetus, followed by a laparotomy. Intraoperative findings revealed peritoneal soiling with gastric contents, herniation of approximately 60% of the stomach into the left hemithorax through a 16 cm diaphragmatic defect, and a perforated gangrenous section on the upper part of the greater curvature of the stomach (Figures 2,3 and 4). The general surgeons performed a two-layer repair of the gastric perforation, while the cardiothoracic surgeons repaired the diaphragmatic rupture (plication) using interrupted horizontal mattress closure with 1 prolene and inserted a chest tube. An abdominal drain was placed, and the wound was subsequently closed.

After the surgery, she was admitted to the intensive care unit (ICU) and placed on ventilatory support with an FiO2 of 50%. She continued to receive intravenous fluids and antibiotics. A post-operative chest X-ray revealed a residual pneumothorax in the left apical region, which was subsequently managed.

She was extubated on postoperative day 4 and began chest and limb physiotherapy. On postoperative day 7, she began having bowel movements, and the nasogastric tube was removed, allowing her to initiate a graded oral intake. The chest tubes and abdominal drains were removed on postoperative day 10, and she was discharged with oral antibiotics and hematinics on postoperative day 11. During a follow-up appointment at the surgical outpatient clinic, she was stable and demonstrating good recovery.



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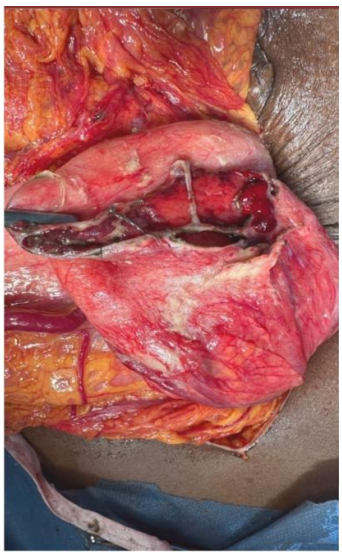

Figure 2 showing the necrotic and perforated area of the stomach

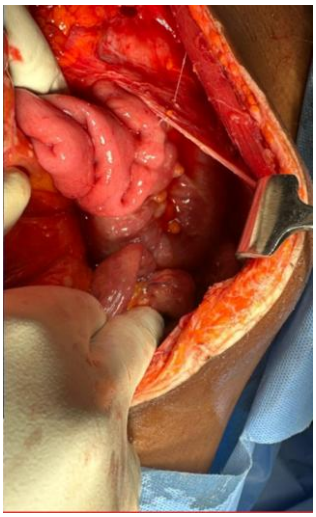

Figure 3 showing bowel herniating into the thoracic cavity

Figure 4 showing the diaphragmatic defect

III. DISCUSSION

Diaphragmatic rupture during pregnancy is a rare but serious condition that can lead to intestinal obstruction, respiratory distress, and even maternal and fetal fatalities[1]. This injury typically results from trauma or a congenital diaphragmatic hernia. Additionally, diaphragmatic eventration can weaken the diaphragm, increasing the risk of rupture due to elevated intra-abdominal pressure during pregnancy. Factors such as repeated vomiting in the first trimester, the growing size of the uterus in the second trimester, and maneuvers like Valsalva or Kristeller during labor contribute to this risk[6].

In our case, repeated episodes of vomiting, violent retches added to a premorbid state of diaphragmatic eventration, may have contributed to the diaphragmatic rupture. This condition is most frequently associated with trauma, whether penetrating or blunt, with an incidence rate reported to be as high as 6%[9]. Diaphragmatic ruptures predominantly occur on the left side in 70 to 90 percent of cases, as observed in our patient. In contrast, right-sided tears are eight times less common than left-sided ones. The abrupt increase in intra-abdominal pressure relative to intrathoracic pressure creates a pressure gradient across the diaphragm. Interestingly, even a violent cough has been documented to lead to traumatic diaphragmatic rupture.

Spontaneous gastric rupture can occasionally be linked to a pre-existing diaphragmatic hernia, as observed in our patient, and it shares similar risk factors with diaphragmatic rupture[10]. There are even reports of it occurring as a late complication of an undetected post-traumatic diaphragmatic rupture. Compromised blood supply due to stomach herniation has been demonstrated to lead to necrosis and subsequent rupture. The lesser curvature is the most common site of rupture, as this area of the stomach is less elastic[2].

It is mostly reported in the third trimester and it is characterized with a significant high number of mortalities. We presented a case involving a multigravida who experienced strangulated herniated viscera during her first trimester. A similar study conducted by Morcello-Lopez et al. examined a 35-year-old primigravida at 18 weeks gestational age, who had a history of left diaphragmatic eventration. This patient suffered both gastric and diaphragmatic ruptures, but ultimately, both mother and child had favorable outcome .[11] while in our case only the mother survived

In general, more cases are observed during the second and third trimesters; however, this differs from our patient's experience, as her symptoms were detected in the first trimester. Chae et al. documented a case involving a 39-year-old woman, G2P1, who was at 26+1 weeks of gestation and experienced strangulation of the entire stomach during pregnancy, yet she and her child had a positive outcome. This condition can occur spontaneously, as seen in our case, or as a result of trauma.

Common symptoms of spontaneous diaphragmatic rupture are pain, nausea, vomiting and dyspnoea.

Gastric perforation often presents with increasing epigastric pain that starts as localized discomfort and may later spread throughout the abdomen. It is important to be aware that this condition can progress to syncope and cardiovascular collapse if not addressed promptly. To improve outcomes, seeking treatment within 12 hours is crucial, as delays beyond this timeframe can significantly raise the risk of fatality. Early intervention can make a meaningful difference in patient prognosis[12]. Diagnosis of diaphragmatic rupture is difficult because of its non-specific symptoms. However, early diagnosis and early reconstructive surgery decrease mortality

A chest X-ray is the most straightforward test for a diagnosis. However, these days, more sensitive tests like magnetic resonance imaging (MRI) or computed tomography (CT) scans are readily accessible in most hospitals, making it easier to get a clearer picture.

Findings indicative of diaphragmatic rupture include an elevated, interrupted, or indistinct hemidiaphragm; the presence of air-fluid levels or bowel loops in the chest; and the presence of a nasogastric tube within the thoracic cavity. In cases of uncertainty, oral contrast studies or a CT scan can be employed for diagnosis. It is crucial to maintain a high level of clinical suspicion in such situations.

Diaphragmatic Eventration is commonly an incidental finding on chest x-ray and subsequent referral is usually made to the surgeons for correction.

Patients close to term and stable are managed conservatively to term before surgery. Indications for surgical intervention for diaphragmatic eventration are the presence of respiratory distress despite medical/conservative management or respiratory failure as seen in our case.

If the diaphragm is not too thin, plication itself gives the diaphragmatic muscle the tautness of a stretched sheet across the torso, acting as a windlass for ventilation

If the plication is not done efficiently, or the muscle is too thin, a mesh may be added to the rim of the diaphragm to strengthen the repair.

There should be a high degree of suspicion of a diaphragmatic rupture especially with background diaphragmatic eventration. Patient should be optimized and have urgent laparotomy for better outcome as delayed surgery increases the risk of fetal and maternal mortality

IV. CONCLUSION

Diaphragmatic rupture in pregnancy is an ever-present risk in patients with previously undiagnosed diaphragmatic eventration. It can occur even with minimal form of trauma such as occurs in common symptoms of early pregnancy itself, frequent vomiting and retching, which can make diagnosis difficult. A high index of suspicion is required if we are to avert serious harm to both the mother and the baby

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