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Bochdalek Diaphragmatic Hernia Complicating Pregnancy in the third Trimester: Case Report

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Abstract

Background: Maternal congenital diaphragmatic hernia complicating pregnancy is an unusual but severe pathology. We only noticed 43 reports on this subject in English between 1959 and 2014.

Case report: A 32-year-old primiparous woman was diagnosed of diaphragmatic hernia at 29 weeks´ gestation and transferred to our hospital. Initially, gastrointestinal symptoms mimicked pathology related to pregnancy, but failure of conservative measures led to performing imaging tests. It was a Bochdalek hernia containing small bowel loops, colon and omentum but not stomach. She was stabilized, but finally cesarean delivery and hernia repair were performed at 32 weeks because of her symptoms, with a successful maternal and fetal outcome.

Conclusion: Obstetricians should take into account these rare digestive disorders for early diagnosis on clinical and radiographic information, especially in pregnant patients who have gastrointestinal symptoms and do not respond to standard treatment. Management of pregnant patients with diaphragmatic hernia is complex. Patients in their third trimester are most commonly managed with cesarean delivery combined with hernia repair, as it is described in our case. If visceral ischemia is suspected, prompt surgery must be performed because delay can result in both fetal and maternal mortality.

Keywords

Bochdalek hernia, Diaphragmatic hernia, Medical and surgical complications of pregnancy, Pregnancy, Treatment

Abbreviations

DH: Diaphragmatic Hernia, CDH: Congenital Diaphragmatic Hernia, MRI: Magnetic Resonance Imaging, CT: Computed Tomography

Introduction

The classification of diaphragmatic hernias (DH) is: acquired, congenital and traumatic. The most common hernias during pregnancy are acquired (hiatal) hernias because of the physiological increased intraabdominal pressure [1].

Most congenital diaphragmatic hernias are diagnosed before birth or in the immediate neonatal period. Bochdalek hernias are the most prevalent CDH in neonates: 1 in 2200 births [2]. Clinical apparition for the first time in the adult period is rare and pregnancies complicated by Bochdalek hernia are even more unusual (to our knowledge 44 cases, including this one, have been reported in the literature since 1928).

However, we don´t know the true prevalence of this kind of hernia in adults. Mullins et al published incidence in adults of 0.17%, based on the 13 138 abdominal computerized tomography (CT) scans reviewed [3]. In their review, 68% of Bochdalek hernias were on the right side, 18% were on the left, and 14% were bilateral. They were found to be more frequent in women than in men (17:5) and Mullins results show that right-sided Bochdalek hernias are more common than we believed up to now in asymptomatic patients (incidental hernias). The left-sided presentation in our patient agrees with the majority of symptomatic cases reported, accounting for 80–90% of all cases [4,5].

When this kind of hernias appear in adult, it is believed that an anatomical failure previously existed, and the hernia is possibly triggered by a trauma or increased abdominal pressure like pregnancy and delivery. In fact, the number of adults with asymptomatic congenital diaphragmatic hernia remains unknown and in women, it may not debut until pregnancy [6]. Symptomatic diaphragmatic hernias during pregnancy, labor and delivery are rare, but the potential consequences are significant. Because of their symptoms simulate other conditions more common in pregnancy, diagnosis and treatment may be delayed, resulting in excessive mortality among these patients [7].

We describe an unusual case of a pregnant patient diagnosed with left-sided Bochdalek hernia in her third trimester that we consider interest publication for obstetrical practice.

Case Presentation

A 32-year-old primiparous woman was admitted to a local hospital (29 weeks´ gestation) because of her gastrointestinal clinic: nausea, vomiting and epigastric pain for three days. She presented normal depositions and no dyspnea. The patient had no medical history of interest. On physical examination, she was afebrile with

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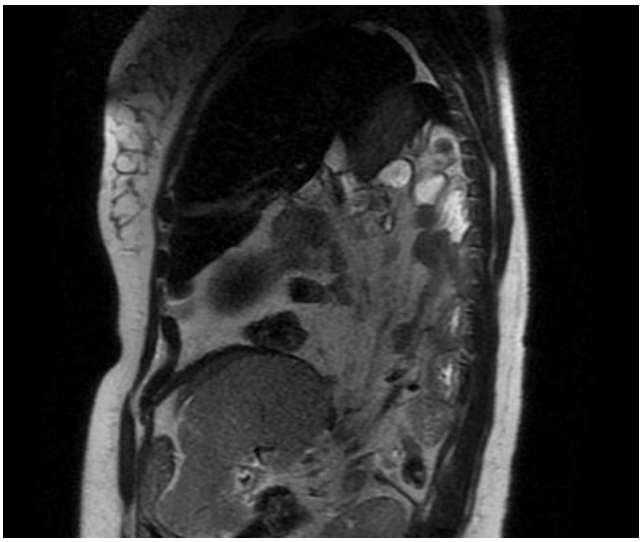

Figure 1: Sagital SSFSE T2-weigthed image, MRI

Posterolateral location of diaphragmatic interruption. Small bowel loops and mesentery are herniated into the thorax. Thickening of anterior fibers of diaphragm.

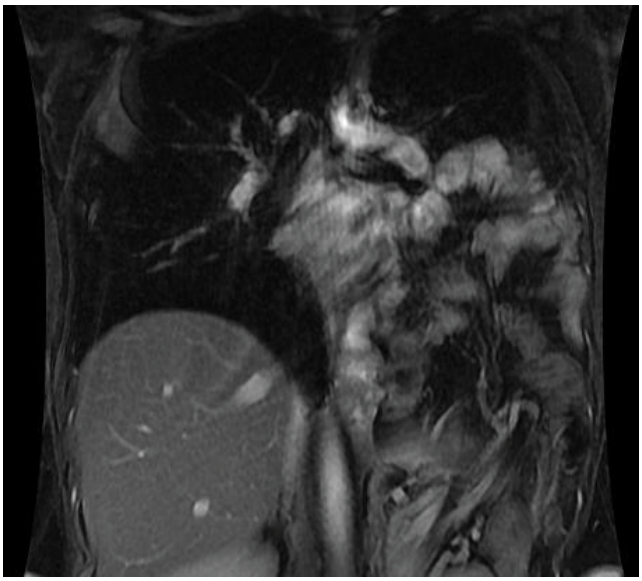

Figure 2: Coronal FIESTA (axial fast imaging employing steady-state acquisition), MRI

Massive herniation of intestinal structures into the thoracic cavity (left-sided diaphragmatic defect of 65 millimeters). No radiological signs of intestinal ischemia. Mesentery thickening and edema. Mediastinal shift to the opposite side.

normal vital signs. Her abdomen was soft, bowel sounds were auscultated without abnormal findings and laboratory tests were correct. There was evidence of fetal wellbeing and there was not uterine activity.

Initially, these symptoms were thought to be due to some pathology related to pregnancy, so she received hyperemesis treatment but no improvement was observed. Then, imaging tests were performed. Maternal abdominal ultrasound was normal but chest X-ray study (anteroposterior and lateral views) showed air fluid levels over the diaphragm and left lung atelectasis. As a DH was suspected, the patient was transferred to our hospital (tertiary care center) for further management. Magnetic resonance imaging (MRI) manifested right and transverse colon and small bowel loops into the left thorax, causing a mediastinal shift. There was a 65 x 44 millimeters posterolateral foramen of Bochdalek defect in left hemidiaphragm but there were no ischemic intestinal signs (Figure 1,2).

Given the stable condition of mother and fetus, we decided to continue expectant management. Two doses of betamethasone were given for fetal maturation. In three days, the patient tolerated soft diet. She was treated with repose, intravenous hydration, metoclopramide and ranitidine. Laboratory tests were carried out serially, without changes. Surgery Service also evaluated her and we agreed to avoid increase in intraabdominal pressure and to plan delivery. She was discharged home seven days after entry, to be controlled twice a week.

The patient was readmitted 12 days later (32 weeks´ gestation). She complained of sudden subesternal and epigastric pain and vomit. Ischemia of some viscera was suspected and so surgery was decided. Emergency cesarean section was performed (Pfannenstiel): healthy child weighing 2230 grams was born. Afterwards, laparotomy through left subcostal incision showed jejunum, ileum, right and transverse colon and omentum in the left hemithorax. The viscera had emerged through 6 x 4 centimeters posterolateral diaphragmatic defect and they were reduced to the abdomen easily. There was no evidence of ischemic organic damage. The defect was sutured with interrupted stitch in single layer (polyglactin) and a polyglactin mesh supported it. Two drainages were placed: a thoracic one into the left hemithorax and another one into left subphrenic space.

The patient´s postoperative recovery elapsed without complications. She was treated with cefazoline and discharged home on postoperative day 10, after she tolerated a regular diet and deposition was correct. The X-ray control image showed left lung fully expanded.

Discussion

We don´t know the true prevalence of this kind of hernia in adults. Clinical apparition for the first time in the adult period is rare and pregnancies complicated by Bochdalek hernia are even more unusual. Symptoms are related to the defect size and the hernia contents and can be respiratory, gastrointestinal or both. Symptoms in adults are usually chronic but they can also present acutely. Gastrointestinal symptoms include: recurrent abdominal pain, abdominal distension, alteration in depositional habit, nausea and vomiting. Respiratory symptoms such as dyspnea and chest pain are less common, but possible. A pregnant patient with diaphragmatic hernia can remain asymptomatic until advancing pregnancy. As pregnancy progresses, abdominal pressure increases and further herniation is produced. Then, gastrointestinal symptoms show up as in our case and there is a higher risk of strangulation. The main life-threatening complications of this pathology are severe dyspnea and visceral strangulation, which may cause maternal or fetus death [8,9].

Diagnosis in pregnant patients is quite difficult because its symptoms are similar to those of some common pathology during pregnancy, so it is essential to take into account these rare disorders. Heartburn, nausea and vomiting are extensive during pregnancy: the failure of usual treatments to improve the symptoms especially in women with advanced pregnancy should guide the physician to more severe gastrointestinal problems [4]. Auscultation of bowel sounds on chest examination is highly indicative of a diaphragmatic hernia. Nevertheless, physical examination is usually not enough for diagnosis [10].

The key test for diagnosis is chest X-ray, showing air fluid levels or gas in the herniated part of the gastrointestinal tract (over diaphragm), as in our patient. Its sensitivity is about 70% [10], but a normal chest X-ray does not exclude Bochdalek hernia in all cases. Computerized tomography and magnetic resonance imaging are techniques to confirm the diagnosis: both have been demonstrated appropriate but probably contrast CT is elective in non-pregnant patients [5]. We selected MRI because of her pregnant status (third trimester). About our images, we should note that stomach was not herniated into the thorax, despite it is one of the organs most frequently involved [8]. It is worth noting that thoracic ultrasonography also has a role, and although it does not have the same diagnostic sensitivity as MRI or CT, actually it can still provide a diagnosis in the majority of cases. There are specific characteristic findings on thoracic ultrasound imaging that suggest the presence of diaphragmatic hernia.

The management of a pregnant patient with symptomatic DH is really a challenge. Gastric decompression (nasogastric tube) may improve clinical condition and delay surgery until transfer to a tertiary center or until corticosteroids are effective. Antenatal corticosteroids should be dispensed if gestational age is between 24 and 34 weeks and surgery can be postponed [11].

Most appropriate treatment depends on the patient´s symptoms and gestational age. Whenever possible these defects should be detected before pregnancy. So, they will be repaired and situations that may increase intraabdominal pressure will be avoided. If Bochdalek hernia is found out in the first or second trimester of pregnancy, elective surgical prenatal repair is advisable although she is asymptomatic. Preferably, surgery should be performed in the second trimester, because at this moment the risk of teratogenesis, preterm birth and miscarriage is lower [9]. Asymptomatic patients in third trimester are recommended expectant management. They should be followed closely and if any sign of obstruction appears, immediate repair should be carried out [8]. Initially, as our patient had hernia symptoms but no signs of strangulation, conservative management was chosen, but finally emergency surgery was necessary [12].

The most appropriate way to end pregnancy in these patients remains unclear. It has been demonstrated that uterine contractions without Valsalva do not increase the intraabdominal pressure and so, they are unlikely to cause hernia incarceration or repaired hernia rupture. Therefore, a patient with a repaired DH can deliver vaginally, without bearing down [1]. But in case of non-repaired DH most authors accept elective caesarean section; hernia repair can be done at the same time but it is not mandatory [8,9]. Recently, other authors have proposed a more conservative action in asymptomatic defects; they argue that vaginal delivery is safe, whenever precautions are taken: planned labor, regional anesthesia and instrumentation to shorten the second stage, and always with the possibility to immediate surgical repair if complications develop [11]. In our case, this option was not possible because of patient´s symptoms.

The best access for surgery of DH occurring on the left side is controversial. Transabdominal approach is most used; it offers good access to herniated organs and so, it should be used when the patient has signs of intestinal ischemia, like in the present case [10,12]. Laparoscopic and thoracoscopic repairs of the left sided Bochdalek hernia have also been performed successfully [2,11]. As our patient was operated in a surgical emergency a laparoscopic intervention was not advisable. In these surgeries it is often necessary to place a mesh in order to support the diaphragmatic defect closure [5].

In the reviewed literature, the overall mortality in adults is around 10-12%. Logically, it is higher in case of emergency surgery (32%) and lower after planned intervention (3%) [11]. About pregnancy, acute complications are more common during third trimester, delivery and early puerperium. Fetal death accounts for 13% of the cases; poor fetal outcomes result from premature labor (24%) and from maternal hypoxia [8]. If symptoms of obstruction appear, delay in surgery worsens prognosis: fetal and maternal mortality rate is up to 50% of these cases [10]. Mortality is caused by maternal respiratory distress, gastrointestinal tract perforation or cardiogenic shock and therefore hypoxia and acidosis for the fetus.

Conclusions

In summary, Bochdalek hernia is an unusual diaphragmatic hernia in adults and symptomatic cases are even rarer. Although rare, DH complicating pregnancy is possible and it is associated with a high rate of morbidity and mortality both maternal and fetal. We want to point out that physicians should consider diagnosis of a DH in pregnant patients who have gastrointestinal symptoms and do not respond to standard treatment. This present case shows the importance of early imaging test in reaching diagnosis. Regardless of gestational age, if there is any suspicion of visceral obstruction, prompt surgery must be performed.

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