CASE REPORT

Acute gastric incarceration from thoracic herniation in pregnancy following laparoscopic antireflux surgery

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SUMMARY

Diaphragmatic hernia is a rare complication in pregnancy which due to misdiagnosis or management delays may be life-threatening. We report a case of a woman in the third trimester of pregnancy who presented with sudden onset of severe epigastric and thoracic pain radiating to the back. Earlier in the index pregnancy, she had undergone laparoscopic antireflux surgery (ARS) for a hiatus hernia because of severe gastro-oesophageal reflux. Owing to increasing epigastric pain a CT scan was carried out which diagnosed wrap disruption with gastric herniation into the thoracic cavity and threatened incarceration. This is, to our knowledge, the first report of severe adverse outcome after ARS during pregnancy, with acute intrathoracic gastric herniation. We recommend the avoidance of ARS in pregnancy, and the need to advise women undergoing ARS of the postoperative risks if pregnancy occurs within a few years of ARS.

BACKGROUND

Diaphragmatic hernia (DH) is a rare complication in pregnancy which, owing to misdiagnosis or management delays may be life-threatening. This case demonstrates the importance of a careful medical history in diagnosing the condition, and raises a debate about the appropriate timing of antireflux surgery (ARS) in relation to pregnancy.

CASE PRESENTATION

A 38-year-old woman (gravida 2, para 1) presented at 31⁺² weeks gestation with sudden onset of severe pain in the epigastrium and in both hypochondria, radiating to the left chest. The pain was constant, and was followed by nausea and vomiting every 10 min. Three days before admission, she had suffered mild upper respiratory symptoms of coughing and sudden severe pain such that she required admission to hospital. In her medical history, she had polycystic ovarian syndrome and infertility. Seven years previously, following in vitro fertilisation she had a triplet pregnancy which was complicated by severe pre-eclampsia and required a caesarean section at 34 weeks gestation. After that pregnancy, she developed essential hypertension requiring antihypertensive therapy. During the index pregnancy, she had a normal oral glucose tolerant test; she was a non-smoker and received antihypertensive treatment.

In addition, she had a 13-year history of gastro-oesophageal reflux due to hiatus hernia with oesophagitis diagnosed gastroscopically. This was

thought to have originated from bulimia in her adolescence. Despite taking proton pump inhibitors daily, her symptoms worsened, and an endoscopy, 10 months earlier, showed a 7 cm hiatus hernia with a 6 cm dilated cardiac sphincter. pH measurement and oesophageal manometry showed pathological reflux with a pH<4. Owing to the long history of severe gastro-oesophageal reflux, ARS with laparoscopic fundoplication was planned.¹ She conceived spontaneously before the surgery could be carried out, but owing to her history of infertility and strong epigastric pain, she decided to proceed with planned surgery which was performed at 6 weeks gestation. During the second trimester, she was diagnosed with intrauterine growth restriction. Accordingly, it was decided that she should deliver by elective caesarean section near term. On physical examination on admission, the patient had stable vital signs, with no fever or dyspnoea. She had epigastric tenderness but no uterine pain.

INVESTIGATIONS

An ECG showed no signs of cardiac ischaemia. The fetal cardiotocograph showed a reassuring fetal heart rate. Abdominal ultrasound showed normal fetal movements and a breech presentation. Although she had a high leucocyte ($18.7 \times 10^9/l$) and neutrophil ($16.8 \times 10^9/l$) count, her cardiac, liver and pancreatic enzymes were normal. Urinalysis showed no proteinuria and an emergency CT scan of her thorax and abdomen revealed complete herniation of the stomach into the left thoracic cavity.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis of a non-smoking, 30-years old, pregnant woman in third trimester, with a known history of hypertension and severe pre-eclampsia who presents with sudden severe epigastric and thoracic pain radiating posteriorly will include the following conditions: severe pre-eclampsia, severe pneumonia or pleurisy, perforated duodenal ulcer, DH, acute myocardial infarction, acute pancreatitis, ruptured aortic aneurism or biliary colic. This woman had a history of previous severe pre-eclampsia requiring emergency caesarean section at 34 weeks gestation and our principal diagnosis was fulminating pre-eclampsia with HELLP syndrome (haemolysis, elevated liver enzymes and low platelets). HELLP syndrome is a variant of pre-eclampsia in which the clinical signs, symptoms and investigations can present before hypertension and proteinuria.² The symptoms of

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Figure 1 Coronal CT image showing wrap disruption with gastric herniation in the left side of the thoracic cavity. Yellow arrows: stomach. Blue arrow: fundoplication wrap.

HELLP are often diffuse with severe nausea, projectile vomiting, pain in the right hypochondrium and epigastrium, as well as a bleeding disorder. In rare cases, the condition results in disseminated intravascular coagulation or eclampsia.³ Women with hypertension have a higher risk of developing pre-eclampsia with or without HELLP.⁴ We also had a suspicion of acute posterior myocardial infarction, because of her known hypertension and insulin resistance. The stable vital signs, normal cardiopulmonary status including normal ECG and normal laboratory tests made us consider other options. The sudden onset of epigastric pain with vomiting together with the fact that she had had an ARS in early pregnancy led us to a diagnosis of DH. A plain thoracic x-ray might have made the diagnosis, but many DHs are misdiagnosed using plain radiography.⁵ In our case, an emergency CT scan was performed because of the suspicion of a possible life-threatening DH with incarceration (figure 1).

TREATMENT

A nasogastric tube was inserted to decompress the expanding hernia, and morphine was given intramuscularly. Antepartum glucocorticoids were also administered with the intention of delaying surgery for 48 h to achieve fetal pulmonary maturation. After 18 h observation, an emergency caesarean section followed by laparotomy was performed because of our suspicion of incarceration/obstruction of the gastric fundus and oesophagus through the diaphragm.

OUTCOME AND FOLLOW-UP

The operation confirmed that most of the stomach and some of the greater omentum had herniated through the fundoplication together with a large oesophageal hiatus hernia. The fundoplication was partly disrupted, but still covered 270° and had been fixated to the diaphragm. The oesophageal hiatus had been sutured distally as part of the primary fundoplication, but a total dehiscence was present. There were early signs of ischaemia of the gastric fundus and necrosis of the greater omentum. The gastric fundus was gently retracted into the abdomen resulting in the return of signs tissue perfusion. The necrotic omentum was resected, and the diaphragmatic defect in the

oesophageal hiatus was repaired with non-absorbable sutures. The postoperative course was without complication for the mother, and she was admitted to the paediatric maternity ward. Her female infant was admitted to the neonatal intensive care unit. She had low birth weight and was treated for respiratory distress syndrome with naloxone and continuous positive airway pressure through an endotracheal tube. Owing to increasing oxygen requirements, intratracheal surfactant was administered. After 5 days, the baby was breathing normally in air and her cerebral ultrasound was normal. After 24 days, mother and daughter were discharged in good health.

DISCUSSION

Classification

DH can be divided into three categories: (1) congenital, caused by defects in the diaphragm arising from faulty embryonic development; (2) traumatic, caused by tears in the diaphragm arising from direct or indirect trauma, for example, road traffic accidents; and (3) acquired, which develop at points of anatomical weakness (hiatus or para-oesophageal hernias).⁵ The prevalence of congenital DH is unknown, although one study using radiological findings estimated the prevalence to be 0.17%.⁶ The prevalence of hiatus hernia is more frequent and increases with age and body mass index, but the correlation with gastro-oesophageal reflux is not always present. One study found a prevalence of hiatus hernia of 19–25% among non-pregnant women with gastro-oesophageal reflux who were scheduled for ARS.⁷ The prevalence of hiatus hernia during pregnancy is unknown, but is estimated to be six times more common than the other two categories.⁸ Up to 85% of pregnant women experience symptoms of gastro-oesophageal reflux⁹ and DH is a rare complication of pregnancy. A recent review of the literature, from 1929 to 2006, reported only 36 cases of DH complicating pregnancy.¹⁰

Mechanism

The enlarging gravid uterus increases intra-abdominal pressure, and increasing progesterone levels cause ligamentous laxity and diaphragmatic muscle relaxation. During delivery, the sharp rise in intra-abdominal pressure pushes the diaphragm up, while the bearing down forces of the Valsalva manoeuvre push the diaphragm down. This results in opposite forces, which can tear the diaphragm at its weakest point, causing herniae in individuals with diaphragmatic defects.^{3 5 11–13} This theory is supported by the fact that the majority of maternal DHs which complicate pregnancy occur during the antenatal period with most of these in the third trimester.

Symptoms

The clinical presentations of maternal DH during pregnancy range from mild epigastric discomfort and nausea, to more severe symptoms with vomiting, upper abdominal and chest pain. The symptoms are often somewhat diffuse, and can be mistaken from normal pregnancy complaints. However, life-threatening complications can occur such as strangulation, obstruction, ischaemia or necrosis of herniated viscera with cardio-respiratory decompensation by the presence of abdominal viscera in the chest.^{10 13} Up to 50% are misdiagnosed because of atypical symptoms and the failure to perform chest radiographs.⁵ Of the 36 reported cases of DH complicating pregnancy, the most common mode of presentation was pain and vomiting (61%), with 32% of the cases presenting with evidence of mediastinal deviation. In the remaining 6%,

respiratory failure was the dominant presenting problem with approximately half presenting during the antepartum period, most often in the third trimester. The remainder presented intrapartum or immediately post partum. Maternal and fetal death occurred in 10 and 9 of the 36 reported cases, respectively. Cases resulting in maternal death were more likely to occur during labour or immediately post partum and were commonly associated with strangulation of the herniated organs.¹⁰

Management of DH in pregnancy

A patient who presents with signs and symptoms of visceral strangulation presents a surgical emergency, and immediate operative intervention is indicated irrespective of fetal maturity. This condition is associated with a high rate of materno-fetal death and, of the 10 maternal deaths reported to date, 7 did not have operative intervention.^{10–14} Patients presenting with symptoms without signs of visceral strangulation, may be managed conservatively until fetal maturity is achieved.¹⁵ Recommendations for the management of women with symptomatic hiatus hernia during pregnancy as well as the timing of ARS in women who wish to conceive, is not well described. ARS with laparoscopic fundoplication is a well-established and successful procedure for treatment of gastro-oesophageal reflux with a success rate of 95%.¹ Symptoms of gastro-oesophageal reflux are experienced by up to 85% of pregnant women⁹ and concerns have been raised that increasing intra-abdominal pressure during pregnancy from vomiting and/or during delivery in women with previous ARS can cause wrap disruption or herniation. Many physicians who manage gastro-oesophageal reflux fear this, and consider ARS contraindicated in women of child-bearing age. In non-pregnant women, the risk of severe adverse events after ARS is low. The quoted rate of intrathoracic gastric migration is 0.17%.¹⁶ Of two small studies on the effect of pregnancy, 2–6 years after ARS, one did not report long-term adverse outcomes or anatomical failure rates,¹⁷ and the other reported one case from 25 women of acute intrathoracic gastric migration at 20 weeks gestation, 6 years after ARS.⁹

CONCLUSIONS

We conclude that the mechanisms leading to this condition were high baseline intra-abdominal pressure from the third trimester uterine size and ligamentous laxity because of high progesterone levels, supplemented with acute rise in intra-abdominal pressure from powerful coughs with contractions of the diaphragm. Together these opposite forces resulted in wrap disruption with complete herniation of the gastric fundus and part of the greater omentum into the thoracic cavity with subsequent ischaemia and necrosis. Our case report is, to our knowledge, the first to describe a severe antepartum adverse effect of ARS performed in early pregnancy with acute wrap disruption and gastric herniation. We recommend ARS during pregnancy should be avoided if at all possible, as well as reflection on the recommended time interval to embarking on a subsequent pregnancy after ARS. The optimal timing of ARS in relation to subsequent pregnancy remains to be evaluated.

Learning points

- ▶ Consider diaphragmatic hernia as a cause, when a pregnant woman with a previous history of antireflux surgery (ARS) presents with sudden severe epigastric pain and vomiting, especially in the antenatal period.
- ▶ A plain thoracoabdominal radiograph with a nasogastric tube in situ can diagnose the condition, but can be misleading. In cases with acute symptoms, resonance CT-scan or MRI is indicated.
- ▶ In cases with suspicion of acute ischaemia, obstruction, strangulation or incarceration of herniated organs, diagnostic procedures should not delay acute explorative laparoscopy/primary laparotomy in the third trimester.
- ▶ Ideally, ARS should not be performed during pregnancy. The optimal timing of ARS in relation to a subsequent pregnancy remains to be evaluated.
- ▶ In cases of recent ARS before pregnancy, one might consider steroid treatment for prophylactic lung maturation from gestational age of 24 weeks.

Competing interests None.

Patient consent Obtained.

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