



Maternal congenital diaphragmatic hernia which first presented in second pregnancy

Sir,

We report a rare case of maternal congenital diaphragmatic hernia (CDH) which first presented in a second pregnancy with epigastric pain. A 17-year-old female, who had a preterm delivery at 36 weeks in her first pregnancy, was admitted with threatened preterm labour at 31 weeks of gestation. She complained of persistent chest and epigastric pain associated with repeated vomiting following the cessation of uterine contractions with tocolysis. Abdominal examination showed a soft and non-irritable uterus with epigastric tenderness, without peritoneal signs. Electrocardiography showed sinus rhythm with no acute ischaemic changes. Both serum creatine kinase and troponin T were normal. The epigastric pain progressed further and became generalised to the whole abdomen. The patient was initially managed as acute pancreatitis in pregnancy, as serum amylase was elevated to 1435 U/L (25–124 U/L). However, chest x ray showed upward displacement of the bowel into the left thoracic cavity, and magnetic resonance imaging confirmed left diaphragmatic hernia with stomach and left-sided bowel loops herniated into the thorax (Fig. 1). Emergency lower segment caesarean section, together with laparotomy for repair of the diaphragmatic herniation, was performed.

CDH is rare, with estimated prevalence of one to four cases per 10,000 live births [1]. The diagnosis of CDH in adults during pregnancy is extremely rare; only approximately 30 cases of CDH which presented

during pregnancy have been reported in English in the past 50 years [2]. The patient's history of an uncomplicated first delivery highlighted the unique feature of this case. In pregnancy, the gravid uterus potentially aggravates the diaphragmatic defect and hernia condition due to increased intra-abdominal pressure. However, the condition remained silent in the study patient's first pregnancy. It is possible that abdominal distension, non-specific abdominal pain, food intolerance with gastro-oesophageal reflux, and intermittent nausea and vomiting were treated as minor ailments of pregnancy. The hernia could have been mild and asymptomatic in the first pregnancy if the defect remained small and the omentum alone was herniated. The hernia defect may have been aggravated further in the second pregnancy, causing bowel herniation. The non-specific symptoms related to CDH mimicking the symptoms of pregnancy could increase diagnostic difficulty. Differential diagnoses including threatened preterm labour, cardiac causes (e.g. aortic dissection, acute coronary event) and acute pancreatitis were considered in this patient. The raised amylase level could indeed be a marker of impending bowel ischaemia. The presence of a bowel shadow in the thoracic cavity on chest x ray is a classical feature of CDH, which should alert clinicians to this rare diagnosis in adults, especially during pregnancy. Incarceration during pregnancy could occur. All symptomatic adult cases of CDH require immediate surgical attention, as bowel incarceration and strangulation are surgical emergencies.

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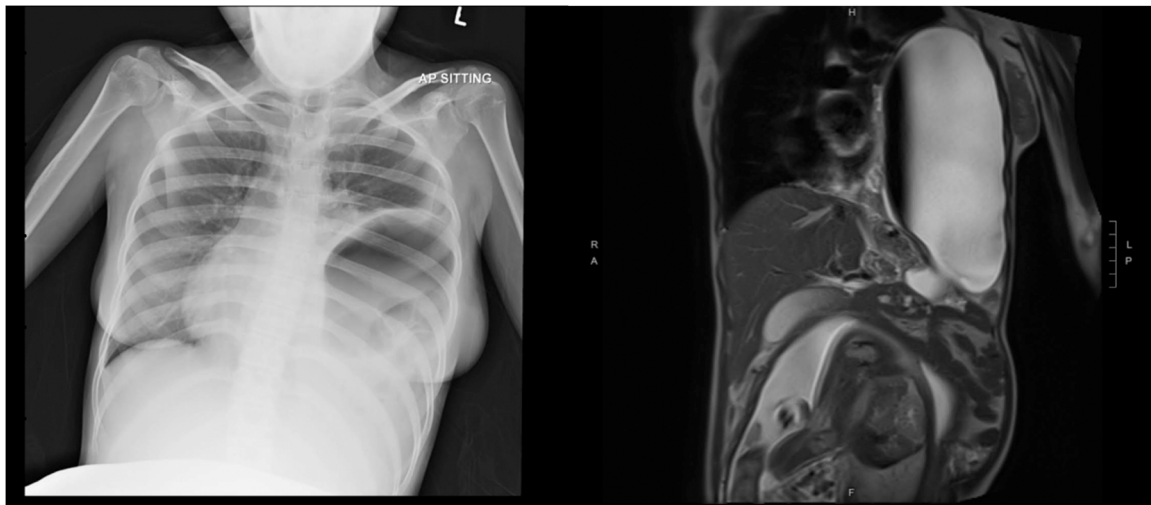


Fig. 1. Chest x ray showing upward displacement of the bowel into the left thoracic cavity, and left diaphragmatic hernia confirmed by magnetic resonance imaging.

Although the atypical presentation of late-presenting CDH in this case led to diagnostic challenge, this clinical entity should be considered in differential diagnoses of patients presenting with unresolved severe gastrointestinal symptoms, especially associated with respiratory distress. Presenting symptoms are non-specific and can be treated as minor ailments of pregnancy. Prompt diagnosis and surgical attention is essential to prevent bowel incarceration.

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Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence

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References

- [1] Burgos CM, Frenckner B. Addressing the hidden mortality in CDH: a population-based study. *J Pediatr Surg* 2017;52:522–5.
- [2] Chen Y, Hou Q, Zhang Z, Zhang J, Xi M. Diaphragmatic hernia during pregnancy: a case report with a review of the literature from the past 50 years. *J Obstet Gynaecol Res* 2011;37:709–14.

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