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CASE REPORT

Maternal Congenital Diaphragmatic Hernia First Manifesting in Pregnancy: A Case Report

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Introduction

Congenital diaphragmatic hernia is a relatively uncommon congenital anomaly, with the diagnosis usually made antenatally by ultrasonography or soon after birth because of respiratory distress in the baby. However, individuals remaining asymptomatic for prolonged periods and only manifesting later have been reported [1]. Congenital diaphragmatic hernia complicating pregnancy is extremely rare, with only 56 cases reported in the literature. The neonatal mortality is reported to be 19% and the maternal mortality 6% [1]. We present a case who presented acutely in the third trimester of pregnancy.

Case Report

Mrs X, primigravida with 32-week gestation presented with severe pain abdomen, backache and vomiting of 1-day duration. She had been admitted to another facility and treated conservatively, but had not improved and was hence referred.

On admission, her vitals were stable except for tachycardia of 120 beats per min. Fetal condition was good as assessed clinically. All initial investigations, including obstetric ultrasound and abdominal ultrasound, were normal. Patient was treated with antacids, antiemetics and analgesics and seemed symptomatically better.

After about 2 h following admission, she developed acute severe breathlessness. Chest examination revealed a dull note on percussion and decreased breath sounds on the left side. X-ray chest showed gas with a fluid level in the left hemithorax which was suggestive of hydropneumothorax (Fig. 1a). Emergency insertion of intercostal drain was done, with escape of gas and some fluid, but the omentum herniated through the drain. A diagnosis of diaphragmatic hernia was confirmed. A nasogastric tube was inserted, which was also noted radiographically to ascend into the left chest (Fig. 1b). At this time, the fetal heart was not detected clinically or by Doppler. The time duration from the onset of acute breathlessness to insertion of the intercostal drain was less than an hour.

Emergency laparotomy was undertaken. A stillborn male baby weighing 1.8 kg was delivered by uterine incision before proceeding to exploration of the diaphragm. A defect of 10 × 8 cm was found in the left posterolateral aspect of the diaphragm, having smooth edges, suggestive of congenital diaphragmatic hernia (Bochdalek) (Fig. 2a). The spleen, stomach, transverse colon and even the sigmoid colon were found to be hypermobile with a tendency to slide through the defect. The defect in the diaphragm was repaired with interrupted non-absorbable sutures. There was also a gastric perforation which was repaired. The surgical repair was undertaken by a team of general surgeons and cardiothoracic surgeon (Fig. 2b).

Postoperative chest X-ray revealed good expansion of both lungs. She withstood the surgery without much alteration of vitals and was managed on ventilatory support postoperatively. However, her vitals deteriorated over the next day, and despite all medical support, she died 25 h

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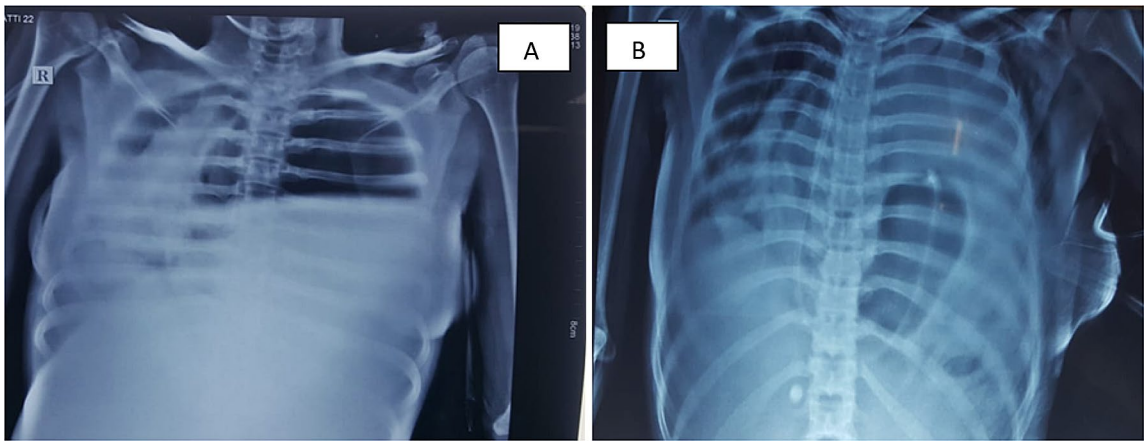

Fig. 1 **a** Chest radiograph showing stomach in left hemithorax, mimicking hydropneumothorax. **b** Ryle's tube seen ascending into left chest (inserted after placement of intercostal drain)

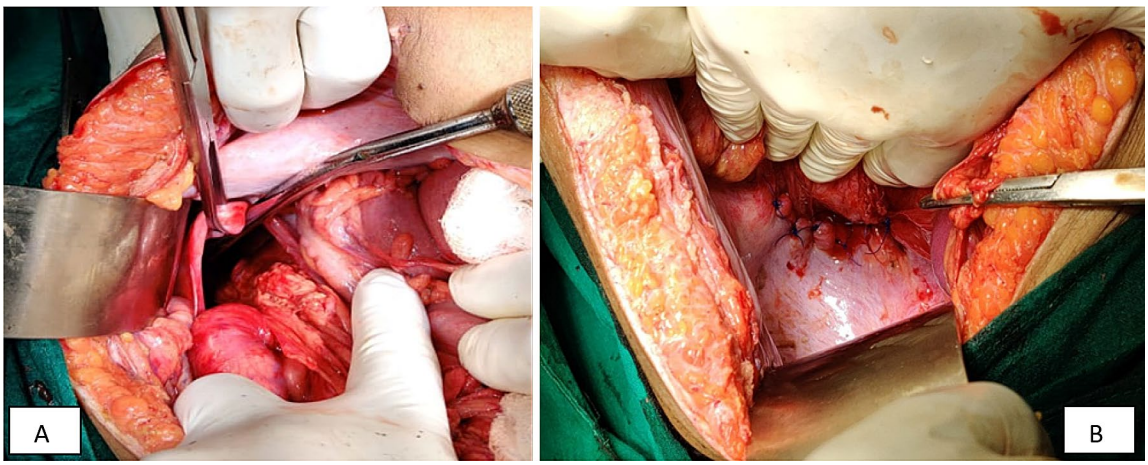

Fig. 2 **a** Left posterolateral diaphragmatic defect with smooth edges. **b** Repaired defect

postoperatively, probably due to severe, irreversible hemodynamic and metabolic insult.

Discussion

Diagnosis of diaphragmatic hernia in late stage of life is not unknown, especially if it is not very big. Pregnancy is a condition in which the intraabdominal pressure is greatly increased and this might lead to decreased ease of the contents sliding in and out of the defect. "Tension gastrothorax" as reported by other authors [2] is the likely cause of acute presentation in this case. Chest X-ray taken in this condition is very likely to be mistaken for hydropneumothorax due to the fluid level in the stomach, and this seems to be a common mistake in the emergency department [3]. The intercostal drain which was inserted for the apparent hydropneumothorax may have caused the gastric perforation in our case, leading to worsening of the condition. This mistake could have been avoided if a nasogastric tube had been inserted before the X-ray examination. Confirming the subdiaphragmatic location of the normal fundal gas shadow, which is seen in 70% of normal chest or abdominal radiographs, is also a good practice to avoid such pitfalls. In other rare presentations, a diaphragmatic hernia may mimic an acute cardiac complication.

Most of the nausea and vomiting of pregnancy is limited to the first trimester, though there are occasional women who continue to experience these symptoms even later in pregnancy. However, women first presenting with these symptoms later in pregnancy should be evaluated critically so as to avoid missing rarer and potentially life-threatening conditions.

Though the reported neonatal and maternal mortality rates in this condition are high, there are reports of successful outcomes in both the mother and the baby [1]. There are also reports of the repair of the defect being undertaken antenatally at a timing remote from Cesarean delivery and also where the mother was subsequently allowed to deliver vaginally [1]. The size of the defect and initial presentation could be the critical factors affecting the decisions and outcomes. In our case, the fetal demise before laparotomy could be because of severe maternal compromise, which further led to deterioration of even the maternal hemodynamic condition and death ultimately.

Ultrasonography, though used widely in pregnancy, has limited application in the diagnosis of maternal diaphragmatic hernia due the enlarged uterus and compromised visibility. The common practice of avoiding X-ray examinations in pregnancy could delay the diagnosis further. The diagnosis could further be complicated by the fact that the contents may or may not have herniated at the time of examination as evidenced by reports where initial chest radiographs appeared benign and repeat examinations revealed the defect [4]. Unless one has a high index of suspicion, the clinical presentation and evaluation findings are likely to be misleading, with potentially adverse maternal and neonatal outcomes.

The repair of a diaphragmatic hernia can be undertaken via laparotomy or thoracotomy. However, it requires a skilled surgical team to undertake this repair. In our case, the repair was undertaken via laparotomy by a team of general surgeons and a cardiothoracic surgeon.

Conclusion

This case has been reported due to its rarity, but primarily to report this manner of presentation and diagnostic challenge. The maternal and neonatal outcomes associated with this condition may be poor because a diagnosis of maternal diaphragmatic hernia is not considered at initial evaluation. This case report highlights the importance of keeping this condition in mind, potentially leading to improved maternal and neonatal outcomes.

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Compliance with Ethical Standards

Conflict of interest The authors declare that there is no conflict of interest of any nature.

Human Participants and/or Animals Rights This is a case report and does not involve research on any human or animal participants.

Informed Consent Consent to report this case was obtained from the maternal aunt of the patient as it could not be obtained from the patient since she succumbed to the disease.

References

1. Reddy M, Kroushev A, Palmer K. Undiagnosed maternal diaphragmatic hernia—a management dilemma. BMC Pregnancy Childbirth. 2018;18:237. https://doi.org/10.1186/s12884-018-1864-4.
2. Næss PA, Wiborg J, Kjellevold K, et al. Tension gastrothorax: acute life-threatening manifestation of late onset congenital diaphragmatic hernia (CDH) in children. Scand J Trauma Resusc Emerg Med. 2015;23:49. https://doi.org/10.1186/s13049-015-0129-8 **PMID: 26104782; PMCID: PMC4477604**.
3. Zefov VN, Almatrooshi MA. Chest X-ray findings in late-onset congenital diaphragmatic hernia, a rare emergency easily misdiagnosed as hydropneumothorax: a case report. J Med Case Rep. 2015;9:291. https://doi.org/10.1186/s13256-015-0755-9 **PMID: 26695937; PMCID: PMC4688985**.
4. Sharma BN, Gupta D. Right sided Bochdalek hernia in an adult: a case report and review of literature. Nepal J Radiol. 2018;8(11):44–7.

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