International Journal of Women's Health

Dovepress

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CASE REPORT

A Rare Case of Diaphragmatic Rupture Due to Ectopic Pregnancy Leading to Haemorrhagic Shock in a Multipara: A Case Report

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Abstract: Abdominal pregnancy is a rare and life-threatening form of ectopic pregnancy. Although the underlying mechanism of this abnormal disorder is unknown, a number of risk factors have been identified, including pelvic inflammatory disease, history of pelvic surgery, intrauterine device use and previous ectopic pregnancy. Diaphragmatic rupture due to ectopic pregnancy is a rare and life-threatening condition that can lead to massive intraperitoneal haemorrhage and haemorrhagic shock. This report presents the case of a 21-year-old woman who presented with 7 weeks and 4 days of amenorrhoea and 14 hours of acute abdominal pain. On examination, she was in haemorrhagic shock with signs of acute abdomen. Emergency exploratory laparotomy revealed a haemoperitoneum of 1500 mL and active bleeding from a 3×2 cm rupture in the right hemidiaphragm. Haemostasis was achieved and the patient recovered well post-operatively. This case highlights the importance of considering diaphragmatic rupture in the differential diagnosis of acute abdomen and haemorrhagic shock, especially in patients with ectopic pregnancy.

Keywords: abdominal pregnancy, ectopic pregnancy, diaphragmatic pregnancy

Introduction

Ectopic pregnancy, defined as the implantation and development of a fertilised ovum outside the uterine cavity, is a potentially life-threatening condition that affects approximately 1–2% of all pregnancies.[1] While the most common site of ectopic implantation is the fallopian tube, accounting for approximately 95% of cases, rare sites such as the cervix, ovary, abdominal cavity and even the diaphragm have been reported.[2]

Diaphragmatic pregnancy, also known as diaphragmatic ectopic pregnancy or subphrenic pregnancy, is an exceptionally rare condition, with an estimated incidence of less than 1 in 10,000 pregnancies.[3] It occurs when the fertilised ovum implants and develops within the muscular diaphragm, typically on the abdominal side. The proposed mechanisms for this unusual location include the retrograde transportation of the fertilised ovum through the fallopian tube into the peritoneal cavity and subsequent implantation on the diaphragmatic surface,[4] the arrest and implantation of the embryo within a congenital or acquired diaphragmatic defect, and the implantation within a diaphragmatic endometriotic lesion that provides a favourable environment for the growth of the trophoblastic tissue.[5]

Regardless of the mechanism, the presence of a developing pregnancy within the diaphragm can lead to severe complications, including rupture, massive haemorrhage and potentially life-threatening haemorrhagic shock.[6] The clinical presentation of diaphragmatic pregnancy can be varied and non-specific, often mimicking other common gynaecological and abdominal conditions. In the early stages, patients may experience symptoms similar to those of a normal intrauterine pregnancy, such as amenorrhoea, nausea and breast tenderness.[7] However, as the pregnancy progresses, complications may arise, including abdominal pain, vaginal bleeding and symptoms related to the compression of adjacent structures, such as dyspnoea, chest pain or shoulder pain.

Received: 16 May 2024
Accepted: 14 September 2024
Published: 1 October 2024

The diagnosis of diaphragmatic pregnancy can be challenging, as the condition is rare and often not suspected initially. Imaging modalities, such as ultrasonography and computed tomography (CT), play a crucial role in the diagnostic process. Ultrasonographic findings may include the presence of a gestational sac or foetal parts outside the uterine cavity, often in close proximity to the diaphragm. The use of CT scans can provide valuable information regarding the location and extent of the ectopic pregnancy, as well as any associated complications, such as haemoperitoneum or organ involvement. However, the definitive diagnosis of diaphragmatic pregnancy involves confirmation by histopathology.

The management of diaphragmatic pregnancy typically involves surgical intervention, with the specific approach depending on the gestational age, the presence of foetal cardiac activity and the extent of complications. In cases of early diagnosis or when foetal demise has occurred, conservative management with surgical removal of the ectopic pregnancy may be considered.[8] However, in cases of advanced gestational age or when significant haemorrhage or rupture has occurred, exploratory laparotomy or laparoscopy may be necessary to control bleeding and remove the ectopic pregnancy.[9]

Diaphragmatic rupture due to ectopic pregnancy is an exceptionally rare but potentially catastrophic complication. The expansion of the gestational sac within the diaphragmatic muscle can lead to erosion and eventual rupture, resulting in massive intra peritoneal hemorrhage and haemorrhagic shock.[10] The clinical presentation in such cases is often dramatic, with acute onset of severe abdominal pain, hypovolaemic shock and signs of acute abdomen. Prompt recognition and timely surgical intervention are crucial in the management of diaphragmatic rupture due to ectopic pregnancy. Failure to promptly control the bleeding and remove the ectopic pregnancy can result in life-threatening consequences, including exsanguination, disseminated intravascular coagulation and multi-organ failure.

Due to its rarity, diaphragmatic pregnancy and its associated complications represent a diagnostic and therapeutic challenge for healthcare providers. Maintaining a high index of suspicion, especially in patients with risk factors for ectopic pregnancy, is crucial for early recognition and appropriate management. Additionally, a multidisciplinary approach involving obstetricians, surgeons and interventional radiologists may be necessary to ensure the best possible outcomes for these complex cases. This case report aims to highlight the importance of recognising diaphragmatic rupture as a potential complication of ectopic pregnancy and emphasise the need for prompt surgical intervention and adequate resuscitative measures in the management of these critically ill patients.

Case Report

A 21-year-old woman (gravida 6, para 1, abortus 4) with a history of one full-term delivery, presented to the emergency department of Yangpu Economic Development Zone Hospital with amenorrhoea for 53 days and acute abdominal pain for 14 hours. Her last menstrual period was on 8 March 2020. According to the patient, 40 days after her last menstrual period, she experienced intermittent vaginal bleeding, which was darker in colour and a lesser quantity compared with her typical menstrual flow. The bleeding persisted for 6 days and then spontaneously stopped.

Fourteen hours prior to presentation, the patient developed severe abdominal pain without any apparent precipitating factors. The pain was described as excruciating and difficult to tolerate, radiating to her right shoulder region. Three hours before arrival, she experienced associated symptoms of dizziness, blurred vision and generalised weakness. She denied any episodes of syncope, nausea or vomiting. Given the severity of her symptoms, she presented to the emergency department, where a posterior vaginal fornix puncture yielded approximately 5 mL of non-clotting, dark red blood. Based on these findings, she was urgently admitted with a provisional diagnosis of a ruptured ectopic pregnancy and haemorrhagic shock.

On initial evaluation, the patient appeared pale and in acute distress. Her vital signs were as follows: pulse rate of 90 beats per minute, respiratory rate of 25 breaths per minute and blood pressure of 70/40 mmHg. Physical examination revealed a distended abdomen with tenderness and rebound tenderness in the lower quadrants. Gynaecological examination revealed normal external genitalia and vagina, with scant white discharge. The cervix was smooth, with cervical motion tenderness. The uterus was anteverted, slightly enlarged and non-tender, and the adnexal regions were not easily palpable.

Laboratory investigations revealed a haemoglobin level of 92 g/L, a markedly elevated white blood cell count of 19.93 × 10^9/L and a positive urine pregnancy test. A CT scan of the abdomen and pelvis showed free fluid in the abdominal and pelvic cavities, suspicious for haemoperitoneum. Based on the clinical presentation, examination findings and laboratory investigations, a provisional diagnosis of haemoperitoneum secondary to a ruptured ectopic pregnancy with haemorrhagic shock was made. The patient was promptly resuscitated with intravenous fluids and blood products, and an emergency exploratory laparotomy was planned.

Intraoperatively, approximately 1500 mL of peritoneal blood was evacuated from the abdominal cavity. The uterus, fallopian tubes and ovaries appeared grossly normal. However, upon further exploration, a 3×2 cm rupture in the right hemidiaphragm was identified, with active bleeding and a clot of approximately 300 g adherent to the rupture site, and a possible diaphragmatic pregnancy was considered. Haemostasis was initially achieved by suturing the bleeding site with 4–0 silk sutures. However, due to the continued oozing and the complicated location of the rupture, the right triangular ligament of the liver was dissected, exposing the diaphragmatic defect and the active bleeding site.

Definitive haemostasis was ultimately achieved by suturing the bleeding site with 4–0 silk sutures and reinforcing the repair with an adjacent segment of the diaphragmatic muscle. The surgical field was irrigated extensively and meticulous haemostasis was ensured before closing the abdomen in layers. During the surgical procedure, the patient received not only six units of packed red blood cells and 600 mL of fresh frozen plasma to compensate for the ongoing blood loss and maintain haemodynamic stability, but crystalloids and colloids to improve volume load. The estimated intraoperative blood loss was approximately 200 mL.

Postoperatively, the patient was closely monitored in the intensive care unit and received analgesics, supportive care and prophylactic antibiotics. Her recovery was uneventful, with gradual improvement in her haemoglobin levels and overall clinical status. The surgical specimen, consisting of the adherent clot and a portion of the diaphragmatic muscle, was sent for histopathological examination. The final pathology report confirmed the presence of chorionic villi, consistent with an ectopic pregnancy implanted within the diaphragmatic muscle. After a hospital stay of 8 days, the patient was discharged in stable condition, with instructions for follow-up and contraceptive counselling to prevent future ectopic pregnancies.

This case highlights the importance of maintaining a high index of suspicion for rare and potentially life-threatening conditions, such as diaphragmatic rupture due to ectopic pregnancy, in patients presenting with acute abdomen and haemorrhagic shock, especially in the presence of risk factors such as a history of ectopic pregnancies or pelvic inflammatory disease.

Discussion

In clinical practice, abdominal pregnancy is a rare and life-threatening form of ectopic pregnancy.[11] It refers to pregnancy in which the fertilised egg or embryo is implanted in the abdominal cavity other than in the fallopian tube, ovary or broad ligament and can be located in the peritoneum, mesentery, omentum, diaphragm or liver. Abdominal pregnancy is divided into primary abdominal pregnancy and secondary abdominal pregnancy, according to the different methods of formation. Secondary abdominal pregnancy is more common, and most are tubal, ovarian or intrauterine pregnancies induced by abortion or rupture. Primary abdominal pregnancy is extremely rare in clinical practice and refers to the pregnancy in which the fertilised egg is directly implanted in the abdominal organs. Although the underlying mechanism of this abnormal disorder is unknown, a number of risk factors have been identified, including pelvic inflammatory disease, history of pelvic surgery, intrauterine device use and previous ectopic pregnancy.[12] Diaphragmatic rupture due to ectopic pregnancy is an exceedingly rare but potentially catastrophic complication. The proposed mechanisms for this unusual implantation site and subsequent rupture include several theories.

One of the most widely accepted hypotheses suggests that the fertilised ovum may undergo retrograde transport through the fallopian tube and into the peritoneal cavity, where it can implant on the diaphragmatic surface. This proposed mechanism is supported by cases where the ectopic pregnancy is found adherent to the diaphragm without any apparent connection to the fallopian tube or ovary; however, the exact route and factors facilitating this retrograde migration remain poorly understood.

The clinical presentation of diaphragmatic rupture due to ectopic pregnancy can be highly varied and non-specific, making the diagnosis challenging, although certain clinical features may raise suspicion for diaphragmatic involvement, such as referred shoulder pain, which is a classic symptom of diaphragmatic irritation. Despite advances in imaging techniques, the definitive diagnosis of diaphragmatic rupture due to ectopic pregnancy is often made intraoperatively, during surgical exploration for suspected ectopic pregnancy or acute abdomen. The intraoperative findings may include the presence of a gestational sac or foetal parts within the diaphragmatic muscle, often accompanied by significant haemoperitoneum and active bleeding.[10]

The management of diaphragmatic rupture due to ectopic pregnancy is primarily surgical and aims to control life-threatening haemorrhage, remove the ectopic pregnancy and repair the diaphragmatic defect. The surgical repair of the diaphragmatic defect may involve direct suturing of the rupture site, while in cases of larger defects, the use of prosthetic mesh or autologous tissue grafts may be necessary.[13] Adequate haemostasis and meticulous surgical technique are crucial to prevent postoperative complications such as haemorrhage, infection or diaphragmatic herniation.[14] In addition, minimally invasive surgery not only reduces surgical trauma and incision-related complications but also reduces recovery and hospital stay.[15]

Diaphragmatic pregnancy is rare in clinical practice. A PubMed search with "diaphragmatic pregnancy" as the keyword found six articles. Among them, Cao[16] and Dennert[17] analysed the important role of human chorionic gonadotrophin combined with B-scan ultrasonography or CT in the diagnosis of diaphragmatic pregnancy from the perspective of imaging examination and emphasised the need to pay attention to upper abdominal pain in menopausal women of childbearing age. Chen,[9] Kang[4] and Wu[18] also emphasised the important role of imaging examination in the diagnosis of abdominal pregnancy and affirmed the value of laparoscopic surgery in the treatment of abdominal pregnancy. Qian et al[14] indicated that ultrasound-guided percutaneous microwave ablation could be crucial for a good outcome in diaphragmatic ectopic pregnancy (Table 1).

In the case reported here, the patient presented with acute abdominal pain, signs of hypovolaemic shock and radiological evidence of haemoperitoneum, prompting an urgent exploratory laparotomy. The intraoperative discovery of the diaphragmatic rupture and the multidisciplinary approach involving obstetricians and general surgeons were critical in achieving successful haemostasis and repair of the defect. The meticulous dissection of the right triangular ligament of the liver and the exposure of the diaphragmatic defect allowed for precise suturing and reinforcement of the repair, ultimately leading to effective haemostasis and a favourable outcome. The postoperative course was closely monitored, with the patient receiving appropriate supportive care, including blood product transfusions, antibiotics and analgesics. The gradual improvement in her haemoglobin levels and overall clinical status reflected the success of the surgical intervention and resuscitative measures.

This case highlights several important lessons and considerations in the management of diaphragmatic rupture due to ectopic pregnancy:

1) In patients presenting with acute abdomen, haemorrhagic shock and risk factors for ectopic pregnancy, healthcare providers should maintain a high index of suspicion for rare and potentially life-threatening conditions, such as diaphragmatic rupture.
2) The management of complex cases, such as diaphragmatic ectopic pregnancies, often requires a coordinated multidisciplinary effort involving obstetricians, general surgeons, interventional radiologists and other specialists as needed.
3) Prompt surgical intervention is crucial in controlling life-threatening haemorrhage and preventing further complications. Delaying surgical exploration in cases of suspected diaphragmatic rupture can lead to catastrophic outcomes.
4) The surgical team must be prepared to adapt their approach based on intraoperative findings and seek additional expertise when necessary, as demonstrated in this case by the involvement of the chief surgeon.

Table 1 Summary of Review Cases Information of the Ectopic Pregnancy Implanted Under the Diaphragm

Author/ Year	Age	The Time of Menopause	The serum β-HCG Level	CT Findings	Surgery	Pathologic Findings
Cao Y 2024[16]	30 years	Menstrual history was regular	13372.08 IU/L	A curved, high-density enhancing mass located beneath the subcapsular hepatic space	Laparoscopic surgery	Embryonic tissue was discerned within the blood clots
Kang OJ 2021[4]	34 years	5 weeks and 6 days	7377.0 IU/L	Approximately 2 cm hypervascular mass in the subphrenic region, with a moderate amount of hemoperitoneum	Laparoscopic surgery	The mass was a product of conception
Wu QL 2021[18]	30 years	1 week and 6 days	13372.08 IU/L	A curved high-density mass beneath the subhepatic space	Laparoscopic surgery	Chorionic villi within the mass, with no features of abnormal trophoblastic proliferation
Qian H 2019[14]	24 years	8 week and 6 days	2526 mIU/mL	There was obviously abnormal density at the top of the right diaphragm	Ultrasound-guided percutaneous microwave ablation	NA
Chen L 2019[9]	33 years	8 weeks	3129.94 IU/L	A 90-mm-long mixed hypodense mass was evident on the upper surface of the right liver lobe	Laparoscopic surgery	Ectopic pregnancy
Dennert IM 2008[17]	34 years	6 weeks	3995 IU/L	NA	Laparoscopic surgery	NA

https://doi.org/10.2147/IJWH.S478590

Conclusion

In conclusion, diaphragmatic rupture due to ectopic pregnancy is a rare but potentially fatal condition that requires prompt recognition and timely surgical intervention. A high index of suspicion is needed for this diagnosis, utilising appropriate diagnostic modalities and implementing a multidisciplinary approach involving experienced surgical teams. Early diagnosis, adequate resuscitation and meticulous surgical technique are crucial in achieving favourable outcomes in these challenging cases. Additionally, this case underscores the need for comprehensive contraceptive counselling to prevent future ectopic pregnancies and reduce the risk of life-threatening complications.

Data Sharing Statement

All data generated or analyzed during this study are included in this published article.

Ethics Approval and Consent to Participate

This study was conducted in accordance with the Declaration of Helsinki and approved for publishing by the ethics committee of Yangpu Economic Development Zone Hospital (2349850Written informed consent was obtained from the participant. The studies were conducted in accordance with the local legislation and institutional requirements.

Consent for Publication

Written informed consent was obtained from the patient for publication of this case report.

Funding

This study did not receive any funding in any form.

Disclosure

All of the authors had no any personal, financial, commercial, or academic conflicts of interest separately.

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