CASE REPORT

Management of splenic ectopic pregnancy presenting with massive haemoperitoneum

Alissa Greenbaum,¹ Richard Miskimins,² Brittany Coffman,³ Jasmeet Paul¹

¹Department of Surgery, University of New Mexico Health Sciences Center, Albuquerque, New Mexico, USA

²Department of Surgery, University of New Mexico, Albuquerque, New Mexico, USA

³Department of Pathology, University of New Mexico Health Sciences Center, Albuquerque, New Mexico, USA

Correspondence to

Dr Alissa Greenbaum, agreenbaum@salud.unm.edu

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SUMMARY

Abdominal ectopic pregnancy (EP) accounts for only 1.3% of EPs and occurs when a fertilised ovum implants in an extrapelvic peritoneal location. Primary splenic pregnancy is a rare type of abdominal EP, with only 16 cases previously reported in the literature. Early diagnosis is essential as delay in treatment carries significant potential for morbidity and mortality. We present the case of a 27-year-old woman presenting with left upper quadrant abdominal pain, elevated human chorionic gonadotropin levels, absence of intrauterine gestational sac and massive haemoperitoneum on transvaginal ultrasound. The patient underwent emergent surgical exploration for high suspicion of ruptured abdominal EP. An open splenectomy was performed when the source of bleeding was confirmed to originate from the left upper quadrant. Final pathology confirmed subcapsular gestational sac implantation within the spleen. While two cases of medical management have been reported, splenectomy remains the current definitive management of rare cases of primary splenic pregnancy.

BACKGROUND

Ectopic pregnancy (EP) is defined as the extrauterine implantation of a fertilised ovum.¹ EP accounts for 2% of all pregnancies, with the fallopian tube being the most common site of implantation (95.5%).^{2–3} Diagnosis is confirmed with elevated β human chorionic gonadotropin (HCG) levels and absence of an intrauterine gestational sac on transvaginal ultrasound. Risk factors for EP include intrauterine devices, smoking, reproductive assistance, specifically in vitro fertilisation procedures with transfer of greater than four embryos, pelvic inflammatory disease, endometriosis, uterotubal malformation, history of tubal surgery and previous EP.^{2–4}

Abdominal EP is a rare and potentially life-threatening event, carrying a mortality rate 7.7 times higher than other types of EP.³ Abdominal EP accounts for only 0.9–1.3% of all EP.^{2–5} Implantation occurs within the peritoneal cavity and is classified as primary or secondary depending on the mechanism of implantation. Secondary is more common and results from continued fetal growth after expulsion from the fallopian tube or other primary site of development. This most often occurs after early rupture of a tubal EP. Primary abdominal pregnancy is the rarest type of EP.³ The most common sites of peritoneal implantation is the pouch of Douglas and posterior uterine wall, though extrapelvic sites may include the greater or lesser omentum, intestines, large vessels, liver and

spleen.^{1–3} Rupture of abdominal EP can lead to haemoperitoneum and haemorrhagic shock.¹ Early and correct diagnosis is crucial, as delay can result in loss of fertility and maternal mortality.⁵

Primary splenic pregnancy is the rarest form of EP abdominally and overall, with only 16 cases previously reported in the literature.^{1–6} The spleen and liver provide especially favourable implantation sites because of their flat surfaces, readily accessible in the peritoneal cavity and extensive vascularity. However, since the placenta cannot attach and develop normally in these solid organs, splenic or hepatic capsular rupture can result, leading to haemorrhage.⁷ The mainstay of treatment after rupture and haemorrhage is splenectomy, though medical management has been purported in certain clinical scenarios. We present the case of a primary splenic pregnancy presenting with massive haemoperitoneum and treated with emergency open splenectomy.

CASE PRESENTATION

A 27-year-old gravida 2, para 2 woman with a history of asthma and hypothyroidism presented to the Emergency Department reporting of sharp left upper quadrant pain of 7 hours duration. Her abdominal pain increased in severity throughout the previous evening, though she initially resisted going to the hospital for medical evaluation. Her gynaecologic history was significant for irregular menstrual cycles and two previous uncomplicated vaginal deliveries. Her last menstrual period ended 3 weeks prior to this newest episode of pain. She agreed to clinical evaluation at the urging of her husband when she developed intractable nausea with vomiting. She reportedly syncope in the hospital elevator on arrival. Vital signs demonstrated tachycardia with a heart rate of 110 bpm, a systolic blood pressure of 98 mm Hg and a temperature of 36.0°C. On examination, she appeared diaphoretic, with diffuse abdominal examination tenderness to palpation though no rigidity or peritoneal signs. The patient was administered intravenous crystalloid fluid resuscitation, antiemetics and opioid pain medication.

INVESTIGATIONS

Serum HCG levels were elevated at 1865 mIU/mL. Serum laboratory studies included a complete blood count, demonstrating a haemoglobin level of 11.4 mg/dL, a white cell count of 13400, platelets of 301 000 and a normal basic metabolic panel except for a mildly elevated glucose level of 114 mg/dL. Liver function tests and international normalised ratio (INR) levels were within normal limits.

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Transvaginal ultrasound revealed a normal uterus measuring 10.5×4.8×8.2 cm, with a 1-mm endometrial stripe, unremarkable fallopian tubes and no intrauterine gestational sac. Abnormal findings included large volume non-simple pelvic free fluid and large volume haemoperitoneum, suspicious for a ruptured EP of unknown location.

DIFFERENTIAL DIAGNOSIS

In the context of elevated HCG levels with no evidence of intrauterine pregnancy, the clinical scenario in this patient's case was best explained by ruptured EP. Additional differential diagnoses for large volume haemoperitoneum and elevated HCG levels with left-sided abdominal pain include ovarian germ cell tumour and gestational trophoblastic disease. Additional considerations would be splenic trauma, rupture of a splenic haemangioma, haemangiosarcoma or splenic artery aneurysm.

TREATMENT

Emergency consult with OB/Gyn was obtained by the emergency department, and she was taken to the operating room for diagnostic laparoscopy. On entering the abdomen, 2 L of haemoperitoneum was encountered and evacuated using a suction irrigator. Thorough inspection of bilateral fallopian tubes and ovaries showed no findings consistent with ruptured EP. A large clot was visualised in the left upper quadrant with a smaller clot seen anterior to the liver. Secondary to findings concerning for pathology in the upper abdomen and a high suspicion of an abdominal pregnancy, the Emergency General Surgery service was consulted to aid in diagnosis and management.

The general surgeon began by placing two additional laparoscopic ports and inspecting the small bowel, from the ileocecal valve towards the ligament of Treitz. The large, organised clot was again noted in the left upper quadrant inferior to the diaphragm and above the spleen. Owing to the size of the clot and an inability to adequately visualise the relevant abdominal structures, the procedure was converted to open exploration through a left subcostal incision. A blood clot measuring ~20 cm was removed and sent for pathologic analysis. Although scant bleeding was noted near the inferior portion and hilum of the spleen, no frank mass or source of haemorrhage was found. A splenectomy was performed after blood continually accumulated in the left upper quadrant and both surgical services expressed a high suspicion for primary splenic pregnancy. No other sources of bleeding were found. After splenectomy, OB/Gyn performed manual vacuum aspiration of the uterus, obtaining good uterine cri with scant amount of tissue removed. These findings were not consistent with intrauterine pregnancy.

OUTCOME AND FOLLOW-UP

Postoperatively she was extubated and monitored in the Trauma Surgical Intensive Care Unit for continued resuscitation. In the morning of postoperative day (POD) 1, she was haemodynamically stable and transferred to the standard hospital ward. Her haemoglobin stabilised at 8.1 mg/dL and human chorionic gonadotropin levels downtrended appropriately to 279 by mIU/mL by 48 hours. On POD3, final pathology confirmed subcapsular ectopic gestation of the spleen (figure 1). The spleen appeared grossly normal, with no areas of apparent infarction or capsular disruption. Sectioning revealed a wedge-shaped, subcapsular pale area measuring 1.1×0.7 cm, containing chorionic villi and implantation site, consistent with ectopic gestation. By POD6, she was tolerating a regular oral diet and ambulating with adequate pain control. She received postsplenectomy meningococcal, pneumococcal and haemophilus influenzae

Figure 1 Histopathologic examination of spleen (×2) demonstrated chorionic villi (bottom left) within normal splenic tissue and adjacent areas of haemorrhage (top right).

vaccinations and was discharged home. At her first clinic follow-up appointment, a repeat pregnancy test was negative and she was recovering well from surgery.

DISCUSSION

Abdominal EP carries a 3% mortality rate, resulting in death ~90 times more often than uterine pregnancy.^{3 8} Abdominal EP presents with large volume haemoperitoneum in greater than one-third of cases.² Risk factors are equivalent to other types of EP. Primary abdominal pregnancy is defined according to criteria published by Studdiford in 1942: (1) normal fallopian tubes and ovaries with no signs of trauma, (2) no evidence of uteroperitoneal fistula and (3) the pregnancy is <12 weeks gestation and involves the peritoneal surface only.^{3 9} Primary splenic pregnancy is especially rare, with only 16 cases previously published.⁶ The average maternal age for splenic pregnancy is 28.7 years, with a gestational age of 7 weeks (range 4–9 weeks), demonstrating a significantly earlier presentation than other types of abdominal EP.^{3 8 10}

Early diagnosis of abdominal EP is crucial. A high level of suspicion should be exercised in a female patient presenting with upper abdominal pain, elevated HCG and absence of intrauterine gestational sac on transvaginal ultrasound.¹¹ Additionally, ruptured abdominal EP should be included in the differential diagnosis of haemoperitoneum in a woman of reproductive age. The decision to obtain further diagnostic studies should depend on the clinical status of the patient. In the case of haemodynamic stability, abdominal ultrasound and CT imaging can be obtained prior to operative treatment.^{1 3 6} While previous authors have not previously employed MRI to aid in diagnosis, this modality may be considered in order to avoid radiation exposure.⁶ If a patient is haemodynamically unstable or has massive haemoperitoneum, as in the case of our patient, emergent surgical exploration is indicated to identify and address the source of active haemorrhage.

Whether diagnosed clinically or through advanced imaging, the mainstay treatment of splenic pregnancy is surgery.^{4 11} Standard for EP operations, a laparoscopic approach may first be attempted in haemodynamically stable patients. In accordance with laparoscopic surgery principles, conversion to an open procedure should be performed if technical difficulty or poor visualisation is encountered. Laparoscopy is converted to an open procedure in 27% of abdominal EP cases.¹ Estimated blood loss during surgery for splenic EP is 1.5 L, and 25% of patients will require blood transfusion.⁸ If ruptured splenic

pregnancy is diagnosed, splenectomy is indicated according to current consensus.^{3 4} This can be performed via either a continued laparoscopic approach or an open incision, either upper midline or left subcostal. If a patient with suspected abdominal EP presents with severe haemodynamic and/or physiologic derangement secondary to haemorrhage, we recommend a midline laparotomy. Similar to a trauma laparotomy, this allows for a damage control operation by packing of all four abdominal quadrants, and stabilising the patient with blood products and fluid resuscitation if needed.

All previously published cases included splenectomy as definitive treatment, with the exception of two cases managed either by laparoscopic or percutaneous injection of methotrexate. Gang *et al*¹ performed laparoscopic enterolysis to expose a 4.3 cm gestational sac of the spleen that was noted preoperatively on abdominal ultrasound and CT imaging. After methotrexate injection of 50 mg/m² directly into the sac, the patient was followed weekly as an outpatient. Two months later, splenic pregnancy was resolved on abdominal ultrasound. Klang *et al*⁶ describe a rare case of splenic EP managed via percutaneous techniques performed by interventional radiologists. The patient, presenting with haemodynamic stability, underwent CT-guided aspiration of the yolk sac, followed by 1 mL injection of methotrexate. HCG levels were persistently high after the procedure, thus 2 mL potassium chloride (KCl) was injected into the gestational sac. A fetal heartbeat, previously seen in the vicinity of the spleen, ceased after this injection. Three months after admission, the splenic pregnancy had resolved per HCG levels and abdominal imaging. The authors mention that while extremely uncommon, one possible complication of this approach is the direct injection of KCl into the maternal circulation with only one known case of cardiac arrest. It should be noted that a percutaneous injection approach does not constitute the current standard of care. It is generally agreed on that successful medical management can occur if pretreatment HCG is <5000 mIU/mL.¹ However, any EP >3.5 cm in diameter, HCG levels >5000 mIU/mL or visible embryo on imaging is more likely to fail medical therapy and should receive surgical intervention.^{5 12}

Primary splenic pregnancy is a rare type of abdominal EP, carrying significant potential for morbidity and mortality. Early diagnosis is essential. In a woman of childbearing age presenting with haemoperitoneum and no tubal evidence of pregnancy, the spleen should be evaluated as a potential site of EP. In haemodynamically stable patients, ultrasound or CT imaging may be obtained prior to surgery to confirm the location of abdominal EP. In unstable patients, concern for rupture and uncontrolled haemorrhage should prompt emergent surgical exploration. Management via direct injection of methotrexate into a splenic gestational sac may be appropriate in a small subset of patients, though this approach has not been widely accepted. Splenectomy, serial HCG levels confirming resolution of pregnancy and postsplenectomy vaccines remain the current recommended management.

Learning points

- ▶ Abdominal ectopic pregnancy (EP) accounts for only 1.3% of all EPs and occurs when a fertilised ovum implants in an extrapelvic peritoneal location.
- ▶ Primary splenic pregnancy is the rare type of EP and if ruptured, it can present with massive haemoperitoneum.
- ▶ A patient presenting with upper abdominal pain, elevated human chorionic gonadotropin levels, absence of an intrauterine gestation and haemoperitoneum should raise suspicion for abdominal or splenic EP.
- ▶ In haemodynamically stable patients with high suspicion for splenic EP, abdominal ultrasound and CT scanning can assist in diagnosis preoperatively.
- ▶ In unstable patients with suspected ruptured splenic pregnancy, emergent surgical exploration and splenectomy is indicated as definitive management.

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