Rare disease

Ruptured heterotopic pregnancy: an unusual presentation of an uncommon clinical problem

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Summary

A 30-year-old nulliparous lady presented to our Emergency Gynaecology Service with a 3-day history of epigastric pain and vomiting at 7 weeks of gestation. An intrauterine pregnancy had been confirmed 3 days earlier when she had attended with an episode of left-iliac fossa pain. Unfortunately, she became more unwell within 1 h of admission and as the cause of her symptoms was unclear, she was taken to the theatre for a joint gynaecology and general surgical diagnostic laparoscopy. This revealed a haemoperitoneum of 2 litres and a ruptured ectopic pregnancy in her left fallopian tube. A left salpingectomy was undertaken to remove the ectopic pregnancy. The patient made an excellent recovery and delivered a healthy baby at 39 weeks of gestation without further complication.

BACKGROUND

Heterotopic pregnancy is an uncommon clinical condition but remains a significant cause of morbidity and occasional mortality. This particular case is important because it highlights (1) that reassurance from the demonstration of an intrauterine pregnancy by ultrasound should not preclude the diagnosis of a heterotopic pregnancy; (2) the importance of considering the diagnosis of a heterotopic or ectopic pregnancy with atypical symptoms, such as epigastric pain and vomiting; and (3) the importance of multidisciplinary team working (gynaecologists with general surgeons) prior to confirmation of the diagnosis.

CASE PRESENTATION

A 30-year-old nulliparous pregnant Caucasian woman presented to our gynaecology service on two occasions. Firstly, she presented with a 24-hour history of mild left iliac fossa discomfort and loose stool. This prompted a visit to our Pregnancy Support Centre, which confirmed a 7-week intrauterine pregnancy by transabdominal and transvaginal ultrasound. Her pain improved so she was discharged back to the care of her general practitioner. Subsequently, she then attended our Emergency Gynaecology Service 3 days later with gradual onset epigastric pain that had become increasingly severe and associated with vomiting. This pain was worse on inspiration and movement, although there was no radiation, including no shoulder tip pain. She had no vaginal bleeding. Her pregnancy had occurred spontaneously without assisted conception and she had no history of pelvic inflammatory disease or previous abdominal surgery. On examination, she appeared pale and distressed. Her heart rate was 92 beats/min with a blood pressure of 128/76 on admission. Her abdomen was soft with severe epigastric tenderness, although there was no rigidity or rebound tenderness. On speculum examination, the cervical os was closed, and the cervix and vagina appeared healthy with a white thick discharge that appeared physiological. On

vaginal examination, the uterus was anteverted with no cervical excitation, no adnexal masses or tenderness.

INVESTIGATIONS

Urinary human chorionic gonadotrophin was positive for pregnancy.

Full blood count revealed a haemoglobin of 97 g/l (decreased but with no previous results to compare with), a white cell count $22.0 \times 10^9/l$ and platelets of $246 \times 10^9/l$. Standard coagulation tests were normal. Her C reactive protein was 12 mg/l. Her renal and liver function tests were normal and amylase was 31 U/l. A group and save was sent for potential blood transfusion in view of this anaemia and pallor.

An emergency abdominal ultrasound (see Figure 1) was reported as follows. *The pancreas was not clearly visualised due to overlying bowel gas but no large pancreatic masses or collections were identified. There was, however, free fluid around the liver and in the abdomen and pelvis. The liver, gallbladder, kidneys and spleen were all unremarkable, and no free gas was visible.*

Figure 1 Abdominal ultrasound scan showing the liver with surrounding free fluid from a ruptured heterotopic pregnancy.

An ECG on admission showed sinus rhythm, 92 beats/min, with normal axis and no ischaemic changes.

DIFFERENTIAL DIAGNOSIS

The principal differential diagnoses included pancreatitis, biliary colic and peptic ulcer disease. However, the previous history of mild left iliac fossa pain raised that possibility of a heterotopic pregnancy (with the knowledge that there was a confirmed intrauterine pregnancy 3 days previously).

TREATMENT

Due to the diagnostic uncertainty, the patient was jointly assessed by the Gynaecology and General Surgery teams. The patient initially declined analgesia, but agreed to 10 mg subcutaneous morphine. She was also given cyclizine and ondansetron for nausea and vomiting. As her heart rate increased from 92 beats/min on admission to 120 beats/min within 1 h, 1 litre of intravenous 0.9% sodium chloride was administered. Over the course of the second hour, her blood pressure decreased from 120/77 to 86/54. She was given intravenous gelofusin and transferred immediately for an emergency diagnostic laparoscopy under general anaesthesia due to the suspicion of intra-abdominal bleeding (the presence of tachycardia, hypotension, anaemia and pallor were suggestive of bleeding from an unknown source). During the laparoscopy, 2 litres of blood was drained from the pelvis, and an enlarged uterus (around 8 weeks of gestation) and a bleeding left tubal ectopic pregnancy (3×4×3 cm) was seen. A left salpingectomy was performed. Haemoglobin fell from 97 to 86 postoperatively and 4 units of red cell concentrate were transfused intraoperatively and postoperatively.

OUTCOME AND FOLLOW-UP

The patient made an uneventful postoperative recovery, had an ultrasound scan the following day that showed a viable intrauterine pregnancy and was then discharged home with oral ferrous sulphate. Histology from the left fallopian tube confirmed a diagnosis of ruptured tubal pregnancy. Follow-up for the ongoing intrauterine pregnancy was arranged in our Pregnancy Support Centre. Further ultrasound scans were undertaken 2 and 4 weeks postoperatively which confirmed viability but demonstrated a haematoma (24 mm) to the left of the uterus. The patient then underwent routine booking and had a normal fetal anomaly scan and an otherwise uneventful pregnancy. The patient delivered a healthy live baby girl at 39 weeks by vaginal delivery following a spontaneous rupture of membranes.

DISCUSSION

We present a case of ruptured heterotopic pregnancy in a lady with no obvious predisposing risk factors. Furthermore, a previous recent ultrasound scan confirmed the presence of an intrauterine pregnancy only. The clinical presentation with epigastric pain and vomiting delayed the diagnosis, which initially prompted investigation for general surgical causes. Despite being physiologically stable during most of the initial assessment, the patient was unwell and the severity of hypovolaemic shock only became apparent when hypotension suddenly developed 2 h after admission. Through prompt multidisciplinary work between general surgery and gynaecology, the patient was rapidly transferred to theatre where the

definitive diagnosis and management for ruptured heterotopic pregnancy was made.

Heterotopic pregnancy is the simultaneous presence of multiple gestations, both intrauterine and extrauterine. The most common extrauterine site is the fallopian tube,¹ although there are less common sites including the cervix² or ovary.³ Even triplet heterotopic pregnancies have been reported.^{4–5} Although rare, the prevalence of heterotopic pregnancy is increasing due to the use of assisted conception technology.^{6–7} However, as in our patient less than 1 in 30 000 pregnancies are affected in those who conceive spontaneously.^{8–9} This was therefore an uncommon diagnosis in our patient who conceived spontaneously. Heterotopic pregnancies have been diagnosed at any stage of pregnancy, between 5 and 34 weeks of gestation,¹⁰ although 70% are diagnosed between 5 and 8 weeks of gestation.⁶ Risk factors for ectopic pregnancy are also applicable to heterotopic pregnancy—these include previous ectopic pregnancy, tubal surgery or pelvic inflammatory disease.¹¹ Our patient had no risk factors which made heterotopic pregnancy a more difficult diagnosis to be made. This highlights that every women of childbearing age with abdominal pain should be considered to have an ectopic pregnancy, regardless of risk factors, until proven otherwise.

The diagnosis of heterotopic pregnancy can be challenging partly because transvaginal ultrasound has a low sensitivity (as low as 26% in one study)¹² and partly because the presentation can be unusual as in our case, where the main symptom was epigastric pain. A recent ultrasound scan in our patient demonstrated an intrauterine pregnancy—this could not provide reassurance of the presence of a fetus in another anatomical location, and clinicians must consider an heterotopic pregnancy despite the ectopic pregnancy not being visualised on the ultrasound scan. It is also possible that ultrasonographers may be less likely to thoroughly visualise for a heterotopic pregnancy if an intrauterine pregnancy is identified because of the rarity of this condition. This poses a problem because delay in diagnosis results in more invasive clinical management and significantly increased risk of morbidity and mortality to both the intrauterine fetus and the mother. In our case, the diagnosis was made after the rupture had occurred. A high index of suspicion is therefore important and the key clinical features for heterotopic pregnancy must be considered: these include the presence of lower abdominal pain, with or without an adnexal mass, peritoneal irritation (in particular shoulder tip pain) and an enlarged uterus.¹³ In our patient, the initial presentation several days previously was with mild left iliac fossa pain (the site of heterotopic pregnancy). However, on presentation to the emergency gynaecological service the main symptoms were epigastric pain and vomiting. This prompted a multidisciplinary approach due to the increased possibility of a general surgical diagnosis, including hepato-pancreato-biliary pathologies. An emergency abdominal ultrasound scan demonstrated free fluid within the abdomen; however, this did not provide a definitive diagnosis, only providing further evidence that significant pathology was present. The early involvement of both general surgery and gynaecology teams meant that when the patient showed physiological signs of worsening hypovolaemia, she was rapidly transferred for a joint diagnostic laparoscopy. During this procedure, 2 litres of blood

was drained from the abdomen and it was deduced that the epigastric pain was due to peritoneal irritation from the haemorrhage of a ruptured heterotopic pregnancy. There are no previous case reports of a ruptured heterotopic pregnancy which present with predominantly epigastric pain and this case report serves as an important lesson of the atypical presenting symptoms for intra-abdominal haemorrhage. It also serves to highlight the remarkable capacity for young adults to compensate physiologically for a significant period of time despite having significant haemorrhage, and then suddenly decompensating when our patient suddenly became hypotensive.

There are a number of management options for heterotopic pregnancy depending on the clinical status of the women, timing of presentation and patient preference. Most women have surgical management. However, there are risks associated with this, in particular the effect of general anaesthesia on both the mother and intrauterine fetus.¹⁴ Some reports suggest that in those undergoing laparotomy, there can be a loss of around 40% of fetuses.¹⁵ There are also other risks of laparotomy over laparoscopy including increased postoperative infection from delayed wound healing. However, laparoscopy with minimal handling of the uterus is considered to be the safest option among most gynaecologists¹⁶—as our patient was very unwell, this laparoscopic salpingectomy was performed. Injection of potassium chloride is increasingly utilised to selective reduce multiple gestations in general obstetrics practice; however, the role for this in reducing an extrauterine heterotopic pregnancy is less clear due to subsequent complications.¹⁷ Expectant management has also been reported,¹⁸ although this is rarely used and requires careful counselling and follow-up. In our patient who was haemodynamically unstable with some diagnostic uncertainty due to the atypical presentation, surgical exploration and management was the best option. In a review of 139 cases where heterotopic pregnancy was managed by surgical intervention, two-thirds of intrauterine pregnancies were born alive.⁶ Our patient successfully delivered a live baby girl at term and this case report serves to highlight the importance of being aware of ectopic and heterotopic pregnancy in any women of childbearing age who presents with any abdominal pain because earlier diagnosis and management will result in improved maternal and fetal outcomes.

Learning points

- ▶ Ectopic pregnancy must be considered in any woman presenting with abdominal pain, independent of risk factors.
- ▶ The presence of a confirmed intrauterine pregnancy on ultrasound does not exclude the presence of an additional pregnancy in another anatomical location.
- ▶ Epigastric pain may be a manifestation of peritoneal irritation and therefore be a sign of intra-abdominal bleeding.
- ▶ Younger adults have a greater capacity to physiologically compensate in response to haemorrhage and this can partially mask how unwell the patient really is.
- ▶ Tachycardia can be an early sign of hypovolaemia and should prompt further assessment for haemorrhage.

Competing interests None.

Patient consent Obtained.

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