CASE REPORT

An unusual cause for epigastric pain in pregnancy. Spontaneous uterine rupture with herniation of the amniotic sac in a 33-week primigravida

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SUMMARY

A 29-year-old in vitro fertilisation patient presented at 33 weeks of gestation with abdominal pain. An abdominal ultrasound revealed a cystic lesion adjacent to the fundus. During caesarean section, a defect at the fundus was identified with herniation of the amniotic sac through this defect. There were no complications postoperatively and the patient made an unremarkable recovery. With at least one maternal death reported in the most recent confidential enquiry into maternal death, uterine rupture is an obstetric emergency and can have catastrophic outcomes. The incidence of uterine rupture as a result of previous perforation is unclear with little published data and few case reports. Cases of uterine rupture after perforation following hysteroscopic resection of fibroids, uterine septum are well published but the authors found no known previous cases related to laparoscopy. Counselling patients post perforation should include discussion regarding the management of future pregnancies and the risk of uterine rupture.

BACKGROUND

Uterine rupture is associated with significant maternal and fetal morbidity and mortality.¹ A systematic review by the WHO found the mean incidence was 5.3/10 000 births.² As the rate of caesarean sections increases worldwide, this is likely to become a more common occurrence. Rupture and/or dehiscence are associated with a scarred uterus—a previous caesarean section scar or myomectomy breaching the uterine cavity—in the majority of cases. Cases have been described, however, where uterine anomalies, an unscarred uterus³ and even cocaine use⁴ have been attributed to this phenomenon.

Uterine rupture typically presents with acute abdominal pain, maternal compromise and/or fetal distress in the presence of augmented or spontaneous labour. The incidence of rupture in an unscarred uterus in the western world, however, is rare and estimated at 0.6/10 000.⁵ A population based cohort study in the Netherlands, a country with one of the lowest caesarean section rates in the western world, found the incidence of uterine rupture to be 5.1/10 000 in women with a uterine scar, and 0.8/10 000 in the unscarred uterus.⁶ As a result, it is not a diagnosis usually associated with primigravid women presenting antenatally with abdominal pain unrelated to trauma.

CASE PRESENTATION

A 29-year-old Indian woman underwent in vitro fertilisation and booked in her first pregnancy at the West Middlesex University Hospital Trust at 11 weeks of gestation. The patient had a past history of endometriosis and had undergone two laparoscopic procedures as part of infertility investigations in the 4-year period prior to conception. With normal booking bloods and anomaly ultrasound she was under joint care with the obstetric and midwifery teams and had an unremarkable first and second trimester.

At 33 weeks of gestation, she presented to hospital with a two-day history of intermittent lower abdominal pain, mainly in the right iliac fossa with radiation to the right loin, dysuria and urinary frequency. A fetal fibronectin test was negative and urinalysis showed minimal proteinuria and leucocytes. There was no sign of fetal distress. A provisional diagnosis of pyelonephritis was made and the patient was started on antibiotics. During the next 24 h of her admission, she continued to have normal fetal monitoring and observations. Her renal function was normal and CRP was less than 5, but a slightly elevated white cell count of 12.4 (neutrophilia 71%) was identified. Over the course of the next 36 h, the patient complained of increasing pain with no relief, despite multiple doses of opiate analgesia. The urine culture was negative and the pain was localised to the right epigastric area as well as the suprapubic area.

INVESTIGATIONS

As the diagnosis was unclear and the patient remained clearly distressed by the pain, a bedside ultrasound scan was performed. This identified a cystic area at the fundus with possible communication to the uterus and amniotic sac. A formal abdominal ultrasound revealed a cystic lesion 8×10 cm adjacent to the uterine fundus (figures 1 and 2). There was a visible defect in the wall of the uterus at this level and the appearances were suggestive of herniation of the amniotic sac through this defect.

On further questioning, the patient had previously been investigated and treated for endometriosis and infertility privately. In 2011 the patient had undergone a laparoscopy and dye procedure and, according to discharge paperwork, there had been no free spill of dye bilaterally but a comment had been made that dye was seen leaking through the uterine fundus. This had not been attributed to

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Figure 1 Transabdominal ultrasound images of the uterine fundus and the defect in the myometrium with herniation of the amniotic sac.

trauma but rather to the consequential high pressures when inserting dye through the Leech Wilkinson cannula. The defect had been sutured with what was described as ‘a laparoscopic technique for haemostasis’.

TREATMENT

Following diagnosis of the herniation, a single intramuscular injection of dexamethasone was administered and an emergency caesarean section was performed at 33+4 weeks gestation after counselling by the paediatric team. A lower segment caesarean section was performed with delivery of a live male infant with Apgar scores of 9 and 10 at 1 and 5 min, respectively, and normal umbilical cord gases. There was no haemoperitoneum and after the uterus was exteriorised the defect at the fundus was identified (figure 3). Membranes filled with amniotic fluid were herniating through the fundus (figure 4). It was not possible to deliver the placenta without draining the herniation first. Once decompressed, the placenta and membranes were delivered intact through the lower segment incision. The 3 cm defect (figure 5) was repaired (figure 6) and the caesarean section completed.

OUTCOME AND FOLLOW-UP

There were no complications postoperatively and the patient made an unremarkable recovery.

Figure 2 Transabdominal ultrasound images of the uterine fundus highlighting the defect in the myometrium.

Figure 3 Herniation of membranes and amniotic fluid through the fundus, seen at caesarean section.

DISCUSSION

With at least one maternal death reported in the most recent Confidential Enquiry into Maternal Death, uterine rupture is an obstetric emergency and can have catastrophic outcomes.¹ As the majority of operative gynaecology becomes ‘minimally invasive’ the use of laparoscopy and hysteroscopy is not only becoming routine but a gold standard for the investigation and treatment of numerous gynaecological complaints. The advantages are well reported including better recovery time, shorter hospital stay and fewer operative complications. Uterine perforation at hysteroscopy has a reported incidence of 1–2.7%.⁷ The incidence of uterine rupture as a result of previous perforation is unclear with little published data and few case reports. A 10-year analysis in India identified a rupture incidence of 1 in 346 pregnancies and of these only one patient was known to have had a previous uterine perforation.⁸ Uccella *et al*⁹ recently reviewed the literature and found 25 reported cases of third trimester prelabour uterine rupture between 1952 and 2011, with 17 of the patients having undergone surgical procedures on the uterus prior to pregnancy. The quoted incidence of rupture in

Figure 4 Herniation of membranes and amniotic fluid through the fundus, seen at caesarean section.

Figure 5 Defect in the uterine fundus after delivery of the placenta.

the unscarred uterus may be associated with undiagnosed perforation after disclosed or undisclosed previous gynaecological surgery but this is hypothetical.

Although uterine rupture after perforation following hysteroscopic resection of fibroids, uterine septum as well as postlaparoscopic myomectomy is well documented, the authors were unable to find any other reports related to laparoscopy and the insertion and use of the Leech Wilkinson cannula. This cannula's use is widespread as part of infertility investigations, both operatively and radiologically, but the authors were unable to find any information as to whether certain, different types of cannulae pose a greater risk of perforation. We suspect that this may indeed be operator dependent.

A theory put forward for the cause of uterine rupture postlaparoscopic myomectomy is deficient healing as a result of poor vascularisation at the uterine incision site.^{4 9} This area would then be predisposed to weakness and possibly rupture. The use of electrosurgery at hysteroscopy and the resulting thermal vascular damage to the myometrium may also result in weakness and subsequent necrosis of the tissue. This may also predispose these areas to higher rates of uterine rupture^{4 9} as in the case above. This may also explain the lack of haemoperitoneum both in our patient and other published case reports postlaparoscopic myomectomy.^{10–12}

Figure 6 The uterine fundus after repair of the defect.

We surmise that the patient had started contracting prematurely and that the increasing uterine pressure resulted in the rupture and subsequent herniation of the amniotic sac. The ultrasound scan images identified that the placenta had implanted at the fundus with the edge abutting the subsequent uterine defect. Had the placenta implanted over the previous perforation site we would argue that the rupture may not have occurred.

Even with prior knowledge of the surgical complications associated with this patient's infertility investigations, uterine rupture of a previously repaired perforation would probably not have been a primary provisional diagnosis. As not all uterine perforations are recognised, this poses a dilemma when counselling patients and acquiring consent from them both for their initial surgery as well as in subsequent pregnancies, as the risk cannot be accurately identified. Our case also highlights the question of management of labour and delivery in women with known uterine perforation, whether repaired or not, as put forward by Tischner *et al.*¹³ In patients who have had their uterine cavity breached by previous surgery, the use of ultrasound to determine the integrity of the uterus and therefore the safety of vaginal delivery has been advocated, and this may be the preferred option in women with a history of significant myometrial injury or surgery.

We propose that counselling patients postperforation should include discussion regarding the management of future pregnancies and the rare but dramatic risk of uterine rupture. This would highlight the importance and need for full disclosure at booking and, in the event of an acute admission, during the antenatal period.

Learning points

- ▶ The importance of thorough and complete history taking from all antenatal patients, not only on acute admission but at all routine booking appointments.
- ▶ All patients who have had previous uterine surgery should see a member of the obstetric team so that appropriate investigation into possible surgical complications can be undertaken.
- ▶ All women with significant myometrial injury must be counselled by a senior member of the obstetric team to ensure the risk of uterine rupture is discussed.

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