CASE REPORT

Advanced abdominal pregnancy in a spontaneous heterotopic pregnancy

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SUMMARY

Spontaneous heterotopic pregnancy is a potentially life-threatening condition rarely considered when a patient with an intrauterine pregnancy is asymptomatic or presents with complaints such as abdominal pain. An advanced abdominal pregnancy as the form of the ectopic component, outside the context of assisted reproduction, is an even more unusual and perilous diagnosis with very few cases reported in the literature. We report such a case in a 34-year-old multipara, with no known risk factors for ectopic pregnancy. The pregnancy was initially misdiagnosed and managed as a dichorionic gestation, complicated with fetal demise of one of the twins. The correct diagnosis was only made after term delivery of the intrauterine pregnancy. The abdominal pregnancy was subsequently managed via laparotomy with no complications.

BACKGROUND

A heterotopic pregnancy implies the simultaneous occurrence of an intrauterine and an ectopic gestation.¹ Its incidence has been estimated to be 1 in 30 000 spontaneous conceptions, but it is higher when assisted reproductive technologies are used.² Abdominal pregnancies account for 1.4% of ectopic gestations and are referred to as advanced when a fetus develops in the mother's peritoneal cavity beyond 20 weeks.^{3,4}

We present a rare combination of two already unusual phenomena: spontaneous heterotopic pregnancy and advanced abdominal pregnancy, in which the intrauterine pregnancy was carried to term with favourable outcome and the abdominal pregnancy was complicated by fetal demise.

CASE PRESENTATION

A 34-year-old woman, gravida 11, para 10, with low social status (class V family according to *Graffar* score)⁵ and records of several child abandonments, was referred to our institution by week 16 of a spontaneous pregnancy. The patient was obese, had type 2 diabetes and was positive for syphilis screening. There was no known history of pelvic inflammatory disease, endometriosis or previous surgeries. She was asymptomatic and had no complaints of vaginal bleeding or abdominal pain prior to the consultation. By that time, a presumed diagnosis of a dichorionic gestation was made based on sex discordance, and standard care was offered.

At 25 weeks' gestation, an ultrasound revealed significant growth discordance between the two

fetuses. Although the first twin had an appropriate growth, the second one had an estimated fetal weight below the 10th centile and oligoamnios. As the umbilical artery Doppler was still in the normal range, a re-evaluation was scheduled in 2 weeks.

The patient deliberately missed several subsequent appointments and exams until 32 weeks when the ultrasound showed normal growth and 8/8 biophysical profile score of the first twin (non-stress test not performed) but no cardiac activity in the second twin. It was also noted that the placenta of the latter had an 'unusual echographic appearance', although no further description or images were provided.

Despite the suggestion of close follow-up, the patient only presented at 37 weeks +6 days when premature rupture of membranes and spontaneous labour occurred, followed by vaginal birth of a male baby, weighing 3170 g with an Apgar score of 9 and 10 at 1 and 5 min, respectively. Since the presenting part of the presumed intrauterine demised twin failed to descend, manual exploration was performed, which failed to identify any intra-uterine content. An ultrasound examination made immediately after showed an empty uterus and suggested an extrauterine intra-abdominal fetus inside an amniotic sac with no cardiac activity. The patient was admitted for postnatal care and additional investigation.

INVESTIGATIONS

A CT scan described a 17×13 cm fetoplacental unit apparently implanted in the fundus and left side of the uterus (figure 1), which was later refuted by an MRI that excluded any attachments of the placenta to the uterus, although it could not indicate safely its implantation site (figure 2).

TREATMENT

The patient initially refused exploratory laparotomy and left the hospital against medical advice 2 days after delivery. Compulsive psychiatric assessment identified oligophrenia, and consent was finally obtained 30 days later. At laparotomy, it was noted that the placenta was attached to the left broad ligament and closely apposed to the left infundibulopelvic ligament. Since the amniotic sac and placenta were clearly individualised, with no invasion of the pelvic sidewall, bowel or mesentery, the excision of the mass was successfully achieved by left annectomy (figure 3). A contralateral tubal ligation was performed for contraception purpose.

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Figure 1 Coronal view of CT in day 1 postpartum showing a 17×13 cm fetoplacental unit apparently implanted in the fundus and left side of the uterus.

There were no intraoperative complications, and the postoperative course was uneventful.

OUTCOME AND FOLLOW-UP

The fetal postmortem examination revealed signs of maceration, and no malformations were identified (figure 4).

The patient is now 4 years remote from surgery with no complications reported.

DISCUSSION

Although still a rare entity in the general population, heterotopic pregnancy's incidence has increased with the widespread use of assisted reproductive technologies. Other risk factors include pelvic inflammatory disease, previous ectopic pregnancy and history of pelvic surgery. The majority of cases are simultaneous intrauterine and tubal pregnancies and commonly present with abdominal pain and vaginal bleeding in the first trimester.² Diagnosis may yet be challenging because the presence of an intrauterine pregnancy falsely reassures most clinicians. The ectopic

Figure 2 Sagittal view of MRI acquired 29 days postpartum showing an intra-abdominal fetus with apparently no connection to the uterus. The correct implantation site could not be determined.

Figure 3 The placenta was implanted in the left broad ligament. The excision was achieved by left annectomy, as it was in intimate relation with the left infundibulopelvic ligament.

component is usually treated surgically, whereas the intrauterine pregnancy is expected to develop normally, despite the higher risk of miscarriage.^{2,6}

Abdominal pregnancies account for 1% of ectopic gestations.^{4,7} A high index of suspicion is needed because the diagnosis is easily missed, with only 20%–40% made preoperatively.⁴ When the abdominal pregnancy is advanced, it has been associated with maternal mortality of 12% and perinatal mortality of 72%.⁸ In this case, the mainstay of treatment is laparotomy, but the timing is controversial as some authors defend immediate surgery, but others delay it until fetal maturity is achieved, when clinically acceptable.⁹ Determination of the placenta's implantation site is of utmost importance since most maternal deaths are a result of uncontrollable bleeding when its removal is attempted. If the extraction is not feasible or safe, some authors suggest leaving the placenta in situ, with or without subsequent administration of methotrexate to hasten its reabsorption. Although it minimises haemorrhage, this approach puts the patient at risk of necrosis, pelvic abscess and wound dehiscence.^{4,10} Abdominal pregnancy brings also complications for the fetus, namely intrauterine growth restriction, fetal demise and various malformations like facial and cranial asymmetry, limb abnormalities or central nervous system defects.^{3,4,11}

Figure 4 Fetal postmortem examination.

Learning points

- ▶ With the widespread of assisted reproduction technologies, heterotopic pregnancy is a more present concern.
- ▶ A high index of suspicion is needed in order to avoid misdiagnosing an advanced abdominal pregnancy as an intrauterine gestation.
- ▶ Management of an intra-abdominal demised fetus is not clearly established, but the risk of developing some complications favours laparotomy and fetal extraction as the primary treatment.

Our patient had no major risk factors for an ectopic pregnancy and was asymptomatic throughout the pregnancy, which hampered the diagnosis. We initially misinterpreted it as a dichorionic intrauterine gestation, an event that is not unusual according to the literature, as the value of ultrasonography alone in the diagnosis of abdominal pregnancies is limited.⁷

The frequency of antenatal care visits was suboptimal during the entire pregnancy, possibly due to low socioeconomic status and oligophrenia. Despite the recommendation of follow-up every 2 weeks after the diagnose of significant growth discordance at 25 weeks, the patient only returned at 32 weeks and no further until premature rupture of membranes and labour onset.

MRI could be more reliable in establishing diagnosis, localising the placenta's implantation site and planning surgery, if the diagnosis was suspected prenatally.⁷ In our case, since the placenta of the abdominal pregnancy did not invade any relevant organs, extraction was safe and uncomplicated.

If the patient continued to refuse laparotomy, we hypothesise that the fetoplacental unit would evolve to a lithopedion ('stone child'), a rare formation that results from the calcification of an untreated abdominal pregnancy. Most of them are incidental findings long after pregnancy during clinical examination, imaging exams for other pathologies or even postmortem.¹² We reinforced the necessity of laparotomy for the theoretical risk of

intestinal obstruction due to adhesions, intra-abdominal abscess and fistula.¹¹

Contributors We state that all authors had significant contribution to this paper and agree with all that is written in it. NM: planning, reporting, conception and design, acquisition and interpretation of data. AFL: planning, conduct, conception and design, acquisition and interpretation of data and reviewing. RC: acquisition and interpretation of data. CP: conduct.

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