

Spontaneous Triple Heterotopic Pregnancy with an Abdominal Pregnancy: The First Case in Literature

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Abstract

Spontaneous triple heterotopic pregnancy is a rare condition involving the coexistence of intrauterine gestation with ectopic gestation. We present the first case of a 29-year-old primiparous woman who consulted for abdominal pain and blackish metrorrhagia. Ultrasound confirmed a viable intrauterine twin pregnancy and an abdominal ectopic pregnancy with a viable embryo. The patient was hospitalized for conservative management of the intrauterine pregnancy and underwent laparotomy for safe excision of the abdominal pregnancy. The intrauterine pregnancy was carried to term without complication, with a scheduled caesarean section at 37 weeks. This case highlights the importance of careful and early evaluation for the diagnosis and management of heterotopic pregnancies, which are particularly complex when involving an abdominal location.

Keywords: Heterotopic Pregnancy; Twin Pregnancy; Abdominal Ectopic Pregnancy; Spontaneous; Laparotomy

Introduction

A heterotopic pregnancy involves the simultaneous occurrence of an intrauterine gestation and an ectopic gestation [1].

Its frequency has been estimated at 1 in 30,000 spontaneous conceptions, but it is higher when medically assisted reproduction techniques are used [2].

Abdominal pregnancies account for 1.4% of ectopic gestations [3,4].

We present a rare combination of two already unusual phenomena: a spontaneous heterotopic pregnancy with an intrauterine twin pregnancy and an abdominal pregnancy, in which the intrauterine twin pregnancy was brought to term with a favourable outcome and the abdominal pregnancy was complicated by the death of the fetus.

Case Presentation

A 29-year-old woman, Gravida 1, para 0, with low socioeconomic status, presented to our institution at the 13th week and a half of a spontaneous pregnancy with abdominal pain and light blackish metrorrhagia. There was no known history of pelvic inflammatory disease, endometriosis or previous surgery.

On examination, she had pain on superficial and deep palpation of the left flank and iliac region. She presented with light blackish bleeding, and abdomino-pelvic examination with vaginal touch revealed no palpable mass, and the cervix was long and closed posteriorly. Haemodynamically, the patient was stable with an arterial pressure at 12/6 and a heart rate at 72 bpm.

Ultrasound confirmed a viable twin dichorionic diamniotic pregnancy, with two live embryos measuring 78 and 81 mm in craniocaudal length at 13 days' gestation and 3 days. A third gestational sac with an embryo measuring 68 mm in length with positive cardiac activity was also visible in the abdominal region (Figure 1). There was minimal effusion in the cul-de-sac of Douglas.



Figure 1: Ultrasound view of a heterotopic pregnancy, twin intrauterine and extrauterine abdominal.

The diagnosis was a threat of abortion in an uncomplicated heterotopic pregnancy, which led to her hospitalization and treatment with tocolytics in the form of progesterone IV delay and antispasmodics such as Spasfon.

Two days later, she underwent a laparotomy which confirmed an abdominal ectopic pregnancy with placenta and amniotic sac enclosed in a shell attached to the left tube and pelvic wall clearly individualized (Figure 2 and 3). The postoperative course was uneventful, and she was discharged home on the fifth day.

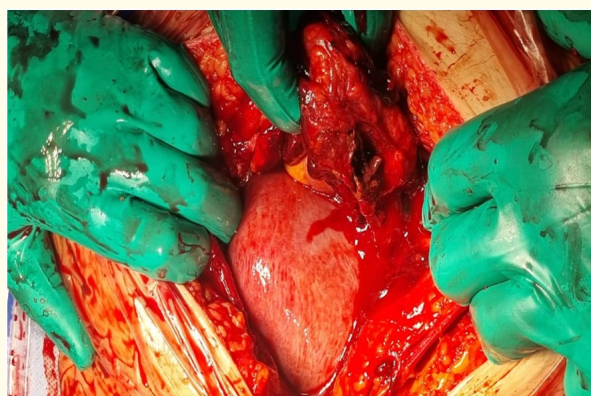




Figure 2: Intraoperative image of extra-abdominal location of pregnancy.



Figure 3: Extra-abdominal pregnancy (after excision).

The pregnancy was monitored in our hospital and proceeded normally until 37 weeks of amenorrhea. A caesarean section was scheduled for the first twin in breech presentation, resulting in the extraction of two newborns, J1 male 2650g Apgar 9/10/10 and J2 female 2540g Apgar 9/10/10.

Discussion

Although still a rare entity in the general population, the incidence of heterotopic pregnancy has increased with the widespread use of medically 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 generally present with abdominal pain and vaginal bleeding in the first trimester [2]. Diagnosis can be difficult, however, because the presence of an intra-uterine

pregnancy falsely reassures most clinicians. The ectopic component is generally treated surgically, whereas the intrauterine pregnancy is expected to develop normally, despite a higher risk of miscarriage [2,5].

Abdominal pregnancies account for 1% of ectopic gestations [4,6]. A high index of suspicion is necessary because the diagnosis is easily missed, with only 20 to 40% of preoperative diagnoses being made [4]. Advanced abdominal pregnancy was associated with a maternal mortality rate of 12% and a perinatal mortality rate of 72% [7]. In this case, the mainstay of treatment is laparotomy, but the timing is controversial as some authors advocate immediate surgical intervention, while others delay it until clinically acceptable fetal maturity has been reached.

The aim of diagnosing heterotopic pregnancy is to detect cases at an early gestational age before rupture or intra-abdominal hemorrhage, which can be achieved through careful patient assessment. Firstly, after diagnosis of the intrauterine pregnancy, the adnexal sites should be assessed for the possibility of heterotopic pregnancy, particularly in cases with risk factors for heterotopic pregnancy. Secondly, early ultrasound evaluation should be carried out with care. However, ultrasound has been reported to have a low sensitivity rate for ectopic/heterotopic pregnancy [8] but it can be improved by systematic and repeated assessments. Certain observations, such as the presence of an ectopic gestational sac with fetal cardiac activity, a fetal ganglion, a hyperechoic ring surrounding the gestational sac and an adnexal mass, indicate a heterotopic tubal pregnancy, with the secondary aim of preserving the intrauterine pregnancy as far as possible.

Determining the site of implantation of the placenta is of paramount importance, as most maternal deaths result from uncontrollable bleeding during attempted extraction. If extraction is not feasible or safe, some authors suggest leaving the placenta in place, with or without subsequent administration of methotrexate to accelerate its resorption. Although this minimizes hemorrhage, it exposes the patient to the risk of necrosis, pelvic abscess and wound dehiscence [4,9]. Abdominal pregnancy also leads to complications for the fetus, including intrauterine growth retardation, fetal death and various malformations such as facial and cranial asymmetry, limb anomalies and central nervous system defects [3,4,10].

In a review of 139 reported heterotopic pregnancies, two heterotopic pregnancies in which the EP was abdominal, removal of the gestational sac and placenta was easily achieved, but in another reported case, during laparotomy, the placental feeding vessel was found to be the ovarian artery and salpingo-oophorectomy was performed [11]. In one case reported by Maciel, *et al.* as the amniotic sac and placenta were clearly individualized, without invasion of the pelvic wall, bowel or mesentery, excision of the mass was successfully achieved by left adnexectomy [12]. In the case of an advanced abdominal pregnancy, with a live fetus, reported by Hailu, *et al.* they preferred to remove the placenta and managed the hemorrhage by packing the area for 24 hours [13]. In another case of full-term abdominal pregnancy with isthmic tubal implantation of the placenta, the placenta was removed by salpingectomy without any attempt to detach it from the fallopian tube [14].

Placental ablation is considered when it is safe and the risk of hemorrhage is low [15]. Consequently, in our case, the placenta was delivered spontaneously and there were no complications.

Our patient had no major risk factors for an ectopic pregnancy, and it was the fact that she presented with symptoms of abdominal pain and the threat of abortion in the form of blackish metrorrhagia coupled with abdomino-pelvic ultrasound that enabled us to make the diagnosis.

MRI could be more reliable for establishing the diagnosis, locating the site of implantation of the placenta and planning surgery [6].

At laparotomy, the placenta and amniotic sac were found to have formed a shell attached to the left tube and the clearly individualized pelvic wall, with no invasion of the bowel or mesentery. Excision of the mass was successfully achieved by left salpingectomy and collapse of the shell (Figure 2).

The available literature on laparoscopic procedures in heterotopic pregnancies is limited overall. Research focusing on the largest number of heterotopic pregnancies (including 10 singleton intrauterine gestations) treated laparoscopically reported three cases of miscarriage and one case of fetal death after chorioamnionitis at 26 weeks [16].

Conclusion

This is a rare case of spontaneous heterotopic pregnancy with abdominal ectopy in which the intrauterine twin pregnancy survives.

All healthcare professionals must bear in mind that intrauterine and extrauterine pregnancies can occur simultaneously. Abdominal ectopy is a serious obstetric condition requiring early diagnosis and prompt treatment. Ultrasound is the diagnostic method of choice, but the existence of an intrauterine pregnancy cannot rule out an ectopic pregnancy, which is why the adnexa must be systematically examined during the first trimester. The life-threatening complication of abdominal ectopic pregnancy is bleeding from the detached placenta. The decision to remove the placenta must therefore be taken on a case-by-case basis.

Conflicts of Interest

The authors declare no conflict of interest.

Author Contributions

All authors contributed to the conduct of this work. All authors also declare that they have read and approved the final version of the manuscript.

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