



Heterotopic tubal pregnancy with a naturally conceived live twin intrauterine pregnancy in a patient with systemic lupus erythematosus: A case report

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ABSTRACT

Heterotopic pregnancy (HP) is defined as the simultaneous occurrence of an intrauterine and extrauterine gestation. Risk factors contributing to this condition are similar to those which contribute to ectopic pregnancy. While a triple heterotopic gestation through natural conception is uncommon, here we present the case of a patient with spontaneous intrauterine twins and a concurrent tubal extrauterine gestation, where the patient also had systemic lupus erythematosus. During the seventh week of gestation, the patient presented with acute abdomen signs and hemodynamic instability; a decision was taken to perform an emergency laparotomy. Haemoperitoneum, a total torsion of the right ovary with salpinx, a ruptured tubal pregnancy and subsequent necrosis were found intraoperatively. The patient was discharged on the sixth postoperative day and monitored throughout her whole pregnancy, with the intrauterine pregnancy progressing uneventfully. Two healthy neonates were delivered by cesarean section at 36 weeks of gestation. In conclusion, physicians treating women of reproductive age should be aware of possible HP, even in the absence of risk factors.

1. Introduction

Spontaneous heterotopic pregnancy (HP) is a highly uncommon medical condition in which extrauterine and intrauterine pregnancies occur simultaneously. This condition is seen in approximately 1/30,000 (1/10000 to 1/50000) of naturally conceived pregnancies, and in approximately 1/100 to 1/3600 pregnancies resulting from in-vitro fertilization (IVF) [1,2]. HP was initially discovered as an autopsy finding by Duverney in 1708 [3–6]. An increase in HP cases has been observed in recent years. This can be attributed to an increased occurrence of pelvic inflammatory disease (PID), increased usage of assisted reproductive techniques (ART) and ovarian stimulation. [3,5,7,8] Following IVF the incidence is nearly 1% of pregnancies.

2. Case Presentation

A 43-year-old pregnant woman (gravida 4, para 3) visited the emergency department of a tertiary hospital with severe abdominal pain and sudden-onset vaginal bleeding. The patient's previous conceptions were natural and she had no history of previous gynecological procedures. The patient had known systemic lupus erythematosus (SLE) with prominent skin rashes around her vulva; the condition was being treated with 5 mg per day of prednisolone. Blood test results on admission were within normal limits, with a blood cell count of 7900 elements/mm³, a haematocrit of 38% and a serum hemoglobin concentration of 12.1 g/dl with a normal blood platelet level (390,000/mm³). The patient's blood pressure was 125/72 mmHg, her heart rate 80 bpm, temperature 36,7 °C, and arterial blood gases were within the normal range (pH 7,42). Her last menstrual period was approximately 7 weeks prior to presentation and a pregnancy test was positive. Abdominal examination

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showed a consistent severe peritoneal pain with rebound detected in the anatomical region of the right ovary.

A vaginal ultrasound scan revealed two intrauterine pregnancies (IUP) with positive fetal heart rate (fetal age of approximately 6 weeks and 4 days) and a large-volume intraperitoneal haemoperitoneum surrounding the swollen right ovary (Fig. 1). The patient was admitted to the intensive care unit (ICU) for further examination and monitoring. Within an hour of admission to the ICU the patient's pain became severe, and she also experienced a decrease in blood pressure (95/50 mmHg) and lowered haematocrit (32%). A decision was made to perform an emergency laparotomy due to hemodynamic instability and acute abdomen.

Total right ovarian torsion with the salpinx, a ruptured tubal heterotopic pregnancy and a subsequent necrosis were found. Estimated blood extravasation was approximately 2500 ml. The necrotic ovary was excised and blood in the peritoneal cavity was removed. Examination of the left ovary revealed an intact corpus luteum, while a vaginal ultrasound scan following surgery showed both IU fetuses with a positive fetal heart rate. An intraoperative transfusion of two units of red blood cells was administered; postoperatively there was no need for an additional transfusion. The patient was discharged on the 6th postoperative day and monitored for the duration of her pregnancy, with the intrauterine pregnancy progressing uneventfully. Two healthy neonates were delivered by cesarean section at 36 weeks of gestation, weighing 2750 g and 2800 g, with Apgar scores of 10 out of 10.

3. Discussion

Heterotopic pregnancy (HP) can occur in various locations, including the abdomen, cervix, cesarean scar, cornu or ovary; however, the most commonly reported location (72%–88.2%) is the fallopian tubes [6,9]. HP presents distinctive challenges in diagnosis and management. Observed complications are nearly 90 times greater than in intrauterine pregnancies, and eight times greater than in tubal ectopic pregnancies [10]. Seventy percent of ectopic pregnancies are diagnosed between five to eight weeks of gestation, 20% are diagnosed between the ninth and tenth week of gestation, while 10% are diagnosed after eleven weeks [11].

Transvaginal ultrasound is essential for the diagnosis of HP and represents the primary diagnostic tool. However, transvaginal ultrasound has low sensitivity and the diagnosis is often missed or overlooked [12–15]. Delayed diagnosis of HP can have severe consequences. An adnexal mass echogenicity helps differentiate a tubal ring of ectopic pregnancy from an ovarian cyst and corpus luteum. Generally, the tubal ring of an ectopic pregnancy is very echogenic compared with ovarian parenchyma. Compared to the ovary, the corpus luteum is often less or



Fig. 1. Two intrauterine pregnancies with positive fetal heart rate, with a fetal age of approximately 6 weeks and 4 days, and a large-volume intraperitoneal haemoperitoneum surrounding the swollen right ovary.

similarly echogenic [16,17]. It is also important to note that the plas-matic b-HCG level is usually difficult to interpret due to the presence of an intrauterine pregnancy (IUP).

Surgical intervention plays a key role in the management of HP [18]. The aim is to remove the ectopic pregnancy while leaving the intra-uterine pregnancy intact [19]. Laparoscopic salpingectomy is the gold-standard surgical approach for HP. Other management options mentioned in the literature include local injection of potassium chloride, hyperosmolar glucose, or methotrexate into the sac under ultrasound guidance, followed by aspiration of the ectopic pregnancy [20].

Pregnancy in the context of systemic lupus erythematosus (SLE) leads to an increased risk of adverse outcomes such as spontaneous abortion, preeclampsia or eclampsia, premature birth and maternal death [21]. Furthermore, there is evidence from a case series of Afro-Caribbean women that maternal SLE is associated with increased incidence of ectopic pregnancy [22]. In this context, further research is needed to investigate a possible association between maternal SLE and ectopic pregnancy.

Herein, we present a rare case of a Caucasian woman with SLE and a spontaneous triple heterotopic pregnancy. Early diagnosis and emergency surgical treatment of the ruptured ectopic pregnancy resulted in an uneventful recovery and the delivery of two healthy neonates by cesarean section at 36 weeks of gestation. Considering SLE as a possible risk factor for ectopic pregnancy, physicians must display a high clinical suspicion regarding ectopic pregnancy in women with SLE.

4. Conclusions

HP has been increasingly diagnosed since the introduction of assisted reproductive technologies, high-resolution ultrasound scans and IVF. There have been cases of heterotopic tubal pregnancies reported which have resulted from clomiphene usage [4]. The probability of an ovarian pregnancy accompanied by IUP comprises 2.3% of all HP cases. Physicians treating women of reproductive age should be aware of possible HP, even in the absence of risk factors. Early diagnosis and management of HP are of great importance and offer an excellent opportunity to have favorable obstetric results for the IUP.

Contributors

Thomas Ntounis contributed to conception and design, was responsible for overall supervision, and drafted the manuscript.

Zacharias Fasoulakis contributed to conception and design, and drafted the manuscript.

Antonios Koutras contributed to conception and design, was responsible for overall supervision, and drafted the manuscript.

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Emmanuel N. Kontomanolis contributed to conception and design, was responsible for overall supervision, and revised the manuscript.

All authors read and approved the final manuscript.

Conflict of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient consent

Obtained.

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