Case Report
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A Rare Case of Ruptured Heterotopic Pregnancy after Natural Conception Demanding Immediate Attention

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Abstract
Objective. We present the case of a ruptured heterotopic pregnancy after natural conception and its management during hospitalization. **Case Report.** The patient presented to the emergency department of our hospital with symptoms of lower abdominal pain and vaginal blood loss. She reported a confirmed intrauterine pregnancy at 8 weeks' gestation following natural conception. The patient was admitted to the hospital with the diagnosis of a ruptured ovarian cyst. Due to hemodynamic instability, an urgent exploratory laparotomy was performed. The histological review showed an ectopic fallopian pregnancy, confirming the final diagnosis of an heterotopic pregnancy. **Conclusion.** Heterotopic pregnancy is a very rare condition, even more so if it happens spontaneously. As a result, it has been insufficiently studied in relation to proper management and timely diagnosis.

Key Words: Eterotopic Pregnancy ▪ Ruptured Adnexa ▪ Pregnancy ▪ Ruptured Cyst ▪ Abdominal Pain.

Introduction

A globally acknowledged complication of Assisted Reproduction Techniques is the phenomenon of ectopic pregnancy. Heterotopic pregnancy is a category of ectopic pregnancy that is present at the same time as an intrauterine pregnancy. Its rate of occurrence in a natural pregnancy is very low, at about 1/10,000 – 1/50,000. Assisted Reproductive Technology elevates this percentage to 1/100 – 1/3600. 1% is not unheard of (1). Generally, well-known risk factors for the existence of an ectopic pregnancy are congenital uterine anomalies (1). Specifically, the embryo transfer of four or more embryos carries a great risk of a heterotopic pregnancy occurring. Distorted anatomy also raises the risk of development (2). The sooner the diagnosis is established, the easier it is to prevent lethal complications. Seventy percent of heterotopic pregnancies are diagnosed during the first 8 weeks' gestation, and the rest during the subsequent 3 weeks (1).

Ectopic pregnancies have the highest chance of presenting in the fallopian tubes, with the cornu coming second. That is also true for heterotopic pregnancies as a result of employing Assisted Reproduction Techniques. The most commonly found risk factors include assisted reproduction, ovarian hyperstimulation, abortion, pelvic inflammatory disease, or previous ectopic gestation with salpingectomy (3). Patients afflicted by an heterotopic pregnancy usually present with nondescript symptoms, such as abdominal pain or pain in the pelvic area. The painful feeling may radiate to the diaphragm or the shoulder, as a result of bleeding occurring in the abdomen. The expulsion of pieces of the decidua may cause spotting (4).

Detecting an heterotopic pregnancy poses several difficulties. Imaging techniques employing contrast agents are usually avoided due to insufficient literature data on the effects on the fetus

(5). An ultrasound scan (either transvaginal or abdominal) used to monitor a normal pregnancy will visualize an intrauterine pregnancy and may fail to show an ectopic one, misrepresenting it as a corpus luteal cyst. Beta human chorionic gonadotropin (B-hcG) levels will rise as in a normal pregnancy, thus causing this diagnostic option to be unreliable (6).

We present the case of a ruptured heterotopic pregnancy after natural conception, and its management during hospitalization.

Case Report

The patient, at 29 years old, presented to the emergency department of our hospital with symptoms of lower abdominal pain and spontaneous vaginal blood loss with blood clots. She reported a confirmed intrauterine pregnancy at 8 weeks' gestation following natural conception. Her obstetric history included a vaginal birth and an abortion for personal reasons. There were no post-abortal complications. Her medical history included hypothyroidism and appendectomy 15 years previously.

The patient was examined for basic clinical signs and they were found to be normal (blood pressure 132/68, heart rate 76 bpm). Vaginal inspection with a speculum showed blood clots with no apparent active hemorrhage. The patient's abdomen was tender on palpation. Blood tests were performed as per standard practice and showed the patient's hematocrit to be 23.9%, prothrombin time/international normalized ratio (PT/INR) 0.94 and she was blood type 0 positive. An ultrasound scan showed an intrauterine pregnancy of Crown-Rump Length: 19,7 mm, gestational age: 9w 2d, FHR: 136 (Figure 1) as well as a cystic formation, 27 × 21 mm, on the left adnexa (Figure 2), and significant free fluid in the pouch of Douglas (Figure 3).

The patient was admitted to the hospital with the diagnosis of ruptured ovarian cyst. As the patient presented with hemodynamic instability while hospitalized (blood pressure 84/46, heart rate 116 bpm), an urgent exploratory laparotomy was performed. During the operation, a significant amount of hemoperitoneum and a ruptured gestation sac on the right fallopian tube were detected. The peritoneal cavity was washed with sterile

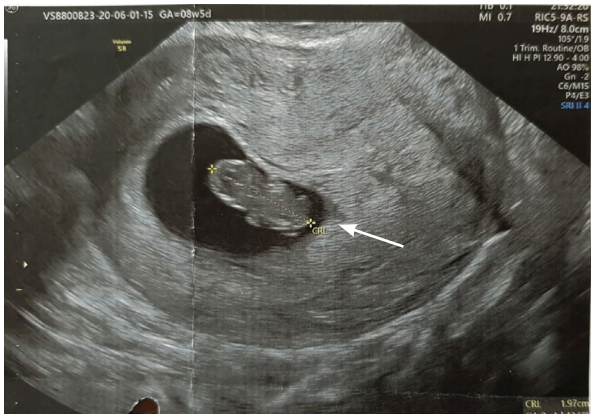

Figure 1. Intrauterine pregnancy Crown-Rump Length: 19.7 mm.

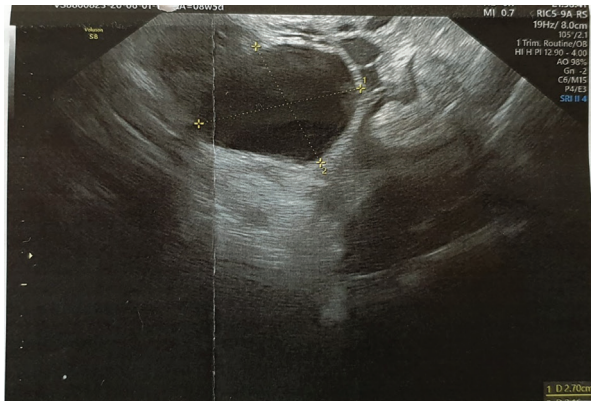

Figure 2. Left adnexa cyst 27 × 21 mm.

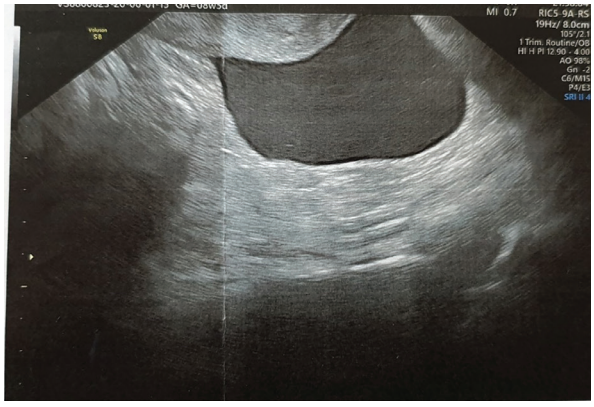

Figure 3. Free fluid in the pouch of Douglas.

saline solution. The right fallopian tube was arrested, ligated and removed alongside the ruptured sac. The uterus and the other adnexa were examined and no abnormalities were found. Complete hemostasis was achieved. A penrose catheter was installed. Intraoperatively, the patient received two blood bags of transfusion. At the end of the operation, a transvaginal ultrasound was performed, and the well-being of the intrauterine pregnancy was ensured.

The post-operative observation was uneventful. The patient was administered IV fluids and broad spectrum antibiotics. Stabilization of the hematoctrit was seen in blood tests. No post-operative complications were detected. No additional transfusion was deemed necessary. Penrose drainage was less than 100mL. The patient was discharged 3 days later in good condition. Prophylactic progesterone support was given, 100 mg × 2 for a week.

A month later, the patient attended for a follow-up examination. An ultrasound scan was performed and no pathological signs or free fluid were found in the peritoneal cavity. The pregnancy continued normally and the patient attended the hospital for a nuchal translucency scan at 12 weeks' gestation. The histological review showed that the sample was an ectopic fallopian pregnancy, confirming the final diagnosis of an heterotopic pregnancy.

Discussion

A natural heterotopic pregnancy is an extremely rare phenomenon. It is common for patients with a ruptured heterotopic pregnancy to be diagnosed during laparotomy, if it is not seen on an ultrasound scan. A gestation in the uterine appendages may be misdiagnosed as an hemorrhagic corpus luteum or an ovarian cyst, if it is noticed at all (7). Spontaneous abortions are a common occurrence to patients with heterotopic pregnancy. The chance for a live birth is 30% lower than in women carrying a normal intrauterine pregnancy. Due to the increased difficulty of the proper diagnosis, there is a great risk of maternal or fetal death (8).

Although difficult, as mentioned, it is possible to detect an heterotopic pregnancy. A mass in the adnexa at the same time as an intrauterine pregnancy gives rise to suspicion of corpus luteum cysts or a heterotopic pregnancy, among other possibilities. The detection of a positive fetal heart rate in the adnexal formation is a sure sign of an ectopic pregnancy. Combining these signs with free fluid in the peritoneal area indicates the diagnosis of a ruptured heterotopic pregnancy (7).

Treatment of heterotopic pregnancy is controversial in the scientific community. The uncommonness of the condition increases the difficulty of shared guidelines in management. Treatment methods that have been used are surgical management by laparotomy or laparoscopy, expectant management, or aspiration of the ectopic sac under sonographic guidance. Aspiration can use drugs that euthanize the embryo in order to reaffirm the corrective course of the ectopic pregnancy (9).

All possible treatments have their positive and negative aspects. Expectant management can avoid the pitfalls of more invasive treatment, however, it is not an option for patients who are hemodynamically unstable or those that present with symptoms, such as abdominal pain or blood loss. For patients in whom expectant management is not an option, surgical treatment is preferred, where physicians have to balance the gain of complete removal of the heterotopic pregnancy with the increased chance of abortion of the intrauterine pregnancy. Another option is aspiration of the ectopic gestation sac under sonographic guidance, possibly with the use of drugs that euthanize the embryo. This is a minimally invasive technique, but highly dependent on the location of the heterotopic embryo, and should be offered as an option only if the the sac is distinctly visualized (10).

Conclusion

Heterotopic pregnancy is a very rare condition, even more so if it happens spontaneously. As a result, it has been insufficiently studied, regarding proper management and timely diagnosis. It is the

authors' belief that the recording of more cases of this phenomenon, as well as further research on preventing its occurrence, will contribute to better understanding and management.

What Is Already Known on This Topic:

A globally acknowledged complication of Assisted Reproduction Techniques is the phenomenon of ectopic pregnancy. Heterotopic pregnancy is a category of ectopic pregnancy that is present at the same time as an intrauterine pregnancy. Its rate of occurrence in a natural pregnancy is very low, at about 1/10,000 – 1/50,000.

What This Study Adds:

We present this case of a ruptured heterotopic pregnancy after natural conception and its management during hospitalization. Heterotopic pregnancy is a very rare condition, even more so if it happens spontaneously. As a result, it has been insufficiently studied, in relation to proper management and timely diagnosis.

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Conflict of Interest: The authors declare that they have no conflict of interest.

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