



Case report

Diagnosis and management of a spontaneous heterotopic pregnancy: Rare case report

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ARTICLE INFO

Keywords:

Heterotopic pregnancy
Intrauterine pregnancy
Ectopic pregnancy

ABSTRACT

Introduction and importance: Heterotopic pregnancy is the occurrence of pregnancies in at least two different implantation sites in the same time. The diagnosis of heterotopic pregnancy remains one of the greatest challenges of the gynecological-obstetrical emergencies.

Case presentation: We report a rare case of spontaneous heterotopic pregnancy of a 32-year-old woman, diagnosed with a heterotopic pregnancy by ultrasound and treated by laparotomy in emergency obstetrical department of Ibn Rochd University Hospital of Casablanca.

Clinical discussion: The existence of intrauterine pregnancy does not exclude an ectopic pregnancy. The occurrence of a spontaneous heterotopic pregnancy without risk factors is a rare event, the clinical symptomatology is often related to a threatened or ongoing abortion, the diagnosis of heterotopic pregnancy is not made until the appearance of signs of hemoperitoneum secondary to a ruptured EP, hence the importance of a systematic ultrasound examination of the adnexa during first trimester ultrasound. The standard treatment is conservative surgery, preferably by laparoscopy. Laparotomy retains its indications especially in forms with hemorrhagic shock. With the aim of preserving intrauterine pregnancy while removing ectopic pregnancy.

Conclusion: The diagnosis of heterotopic pregnancy should not be excluded by the discovery of a UGI in a spontaneous cycle. Diagnosis is often difficult and management should be initiated as soon as possible given the risk of maternal mortality.

1. Introduction

Heterotopic pregnancy (HP) is the occurrence of an intrauterine pregnancy (IUP) and an ectopic pregnancy (EP) simultaneously, whatever its location [1]. The most common ectopic pregnancies are located in the fallopian tubes. Abdominal location increases the risk of maternal mortality up to 90 times higher than a normal IUP [2]. It's a challenge to make the diagnosis of a heterotopic pregnancy. The main treatment consists of removing the ectopic pregnancy, while preserving the intrauterine one. We report a rare case of spontaneous heterotopic pregnancy, complicated by a large hemoperitoneum, with favorable intrauterine pregnancy's evolution after surgery. This work has been reported with respect to the SCARE 2020 criteria [3].

2. Case report

A 32-years-old women, third gesture third pare, mother of two children by vaginal delivery, the patient had no risk factor for heterotopic pregnancy, no notion of hormonal stimulation and no pathological history or similar family cases. She has consulted for acute pelvic pain, associated to a two months menstruation's delay.

In admission, the patient has presented a hypotension of 80/50 mm Hg, a tachycardia of 110 beats/min and a generalized cutaneous-mucosal paleness. However, she complained left iliac fossa pain, and a minimal endocervical bleeding. Furthermore, the uterus was increased slightly in size. The patient was rapidly and urgently conditioned with two large calibre venous route and vascular filling with macromolecules.

Suprapubic and transvaginal ultrasound a live intrauterine pregnancy, the crown rump length was measured at 26.4 mm, which

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<https://doi.org/10.1016/j.ijscr.2021.106184>

Received 19 June 2021; Received in revised form 6 July 2021; Accepted 6 July 2021

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corresponds to 9 weeks and 2 days of pregnancy. Moreover, an adnexal mass has been visualized, in favor of a live ectopic pregnancy, the crown rump length was measured at 17.4 mm, which correlates to 8 weeks of pregnancy, with a large free fluid in the cul-de-sac, the Morison pouch, and the sub-phrenic space. (Figs.1, 2, 3, 4). The hemoglobin level was 8.7 g/dl, β -hCG level of 145.754 IU/ml. The diagnosis of ruptured ectopic pregnancy was strongly suspected.

In view of the state of hemorrhagic shock an urgent laparotomy was therefore indicated under general anesthesia and in dorsal decubitus, it revealed a hemoperitoneum of 700 ml, a ruptured left ectopic pregnancy, and a damaged fallopian tube, that could not be preserved. However, the uterus was slightly increased in size, gravid in appearance (Fig. 5). A left salpingectomy was performed (Fig. 6).

The postoperative period hasn't reveal any complications. Then, progesterone vaginal was administered at a rate of 200 mg 3 times a day. The patient was declared discharged on postoperative day 5.

Two weeks later, ultrasound monitoring showed a normal evolution of the intrauterine pregnancy. The histopathological report confirmed the diagnosis of ectopic pregnancy. At 37 weeks and 5 days, she delivered by spontaneous vaginal delivery.

3. Discussion

Heterotopic pregnancy is a rare form, which is defined by the coexistence of an ectopic and intrauterine pregnancy. The incidence of heterotopic pregnancy is estimated at 1/30.00, above 1/100 when associated with in vitro fertilization, and 1/900 when using clomiphene citrate [4]. The important risk factors for the development of a heterotopic pregnancy include family history, endometriosis, tubal disease, history of pelvic inflammation, high hormone levels, embryo transfer technique [5].

The diagnosis of heterotopic pregnancy remains one of the greatest challenges of the gynecological-obstetrical emergencies. It is often delayed due to the early visualization of an intrauterine sac, with late detection of adnexal abnormalities; the clinical symptomatology is often related to a threatened or ongoing abortion, so an intrauterine pregnancy should not exclude a simultaneous ectopic pregnancy. A detailed history and physical examination are importance to explore all risk factors related to heterotopic pregnancy, which are common to those of EP: unnoticed chronic Chlamydia trachomatis infections. Pelvic inflammatory disease, previous ectopic pregnancies, tubo-ovarian abscess, previous tubal surgery. [6]



Fig. 1. Transvaginal ultrasound: Heterotopic pregnancy, double gestational sac in the uterus and fallopian tube.

Combined pregnancy can result from simultaneous or delayed fertilization. In a literature review, over the period from January 1994 to December 2004, 13 cases of heterotopic pregnancies were spontaneous, and almost 74% were diagnosed early, between 5 and 8 weeks of gestation. However, one case was recognized at 20 weeks [7].

The most common symptoms include abdominal pain, vaginal bleeding, adnexal mass, peritoneal irritation and uterine enlargement [7,8]. If unrecognized, the evolution can be towards hemoperitoneum following rupture of the EP or even maternal shock [9].

The first-line examination is suprapubic and transvaginal ultrasound, which allows the diagnosis of both pregnancies, specifying the vitality of the intrauterine pregnancy and the site of the ectopic pregnancy [10]. Although, the sensitivity of ultrasound can vary from 26.3% to 92.4% [11]. The presence of an intrauterine pregnancy leads to difficulties of interpretation, especially in the youngest pregnancies, when the ovarian pregnancy could be mistaken for a corpus luteum. As the above reported clinical case, both intrauterine and ectopic pregnancies were visualized by ultrasound.

β -hCG's level is not useful for of heterotopic pregnancy's diagnosis. Intrauterine pregnancy masks all underlying β -hCG changes from ectopic pregnancy and vice versa [12].

Treatment can be medical or surgical. It has to be as early as possible. It aims to conserve the UGI, while removing the ectopic pregnancy, preserve the patient's fertility and avoid recurrence. For asymptomatic or hemodynamically stable patients, expectant management can be suggested in order to avoid surgery and transvaginal ultrasound guided aspiration's complications [11]. In case of hemodynamic instability, with signs of EP's rupture, emergency surgery is highly recommended [12]. It mainly involves salpingectomy, salpingotomy or oophorectomy. In some difficult cases it may also require a hysterectomy. Manipulation of the uterus should be minimal, in the order to preserve the UGI [8]. Single incision laparoscopy was first introduced for the treatment of ectopic pregnancy by Ghezzi et al. [13]. Laparoscopy has the advantage of avoiding the risk of uterine manipulation and desiccation, compared to laparotomy, which can cause uterine irritability and postoperative spontaneous abortion. Laparotomy is indicated in cases of hemodynamic instability or large hemoperitoneum.

Due to its teratogenic effect, Intra-muscular injection of Methotrexate may be an alternative only if the UGI is non progressive, or non-viable. It should be noted that a higher rate of abortion of the intrauterine pregnancy has been documented in patients under medical treatment compared to surgical management with respective proportions of 50% and 13% [14].

A majority of women deliver normally at term, with a caesarean rate of 20% due to the presence of two surgically treated corneal pregnancies [15].

4. Conclusion

The frequency of heterotopic pregnancy has increased in recent years, with the emergence of medically assisted procreation. However, its incidence in the spontaneous cycle remains rare and can be life-threatening for the patient. Diagnosis is often difficult. HP should be included in the differential diagnosis of acute abdomen. The standard treatment is conservative surgery, preferably by laparoscopy. However, treatment by laparotomy is not uncommon. Through this case report, we brought to light the importance of exploration, through the patient's history, the physical examination, and all risk factors linked to heterotopic pregnancy in order to make the diagnosis as early as possible.

Provenance and peer review

Not commissioned, externally peer-reviewed.

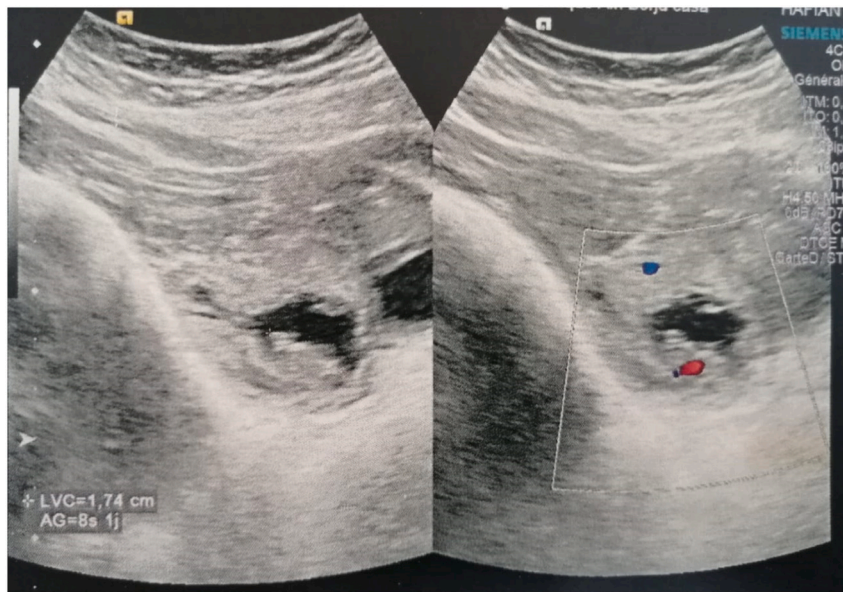


Fig. 2. Pelvic ultrasonography: Ectopic pregnancy in the left fallopian tube.

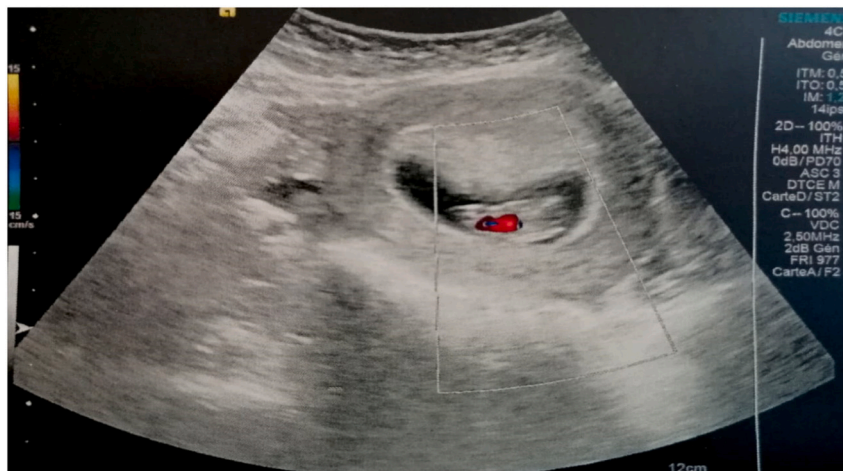


Fig. 3. Pelvic ultrasonography: live intrauterine pregnancy.



Fig. 4. Abdominal ultrasound: hemoperitoneum in the Morison pouch.



Fig. 5. Intraoperative finding of ectopic pregnancy and hemoperitoneum.



Fig. 6. Salpingectomy, trophoblast and embryo.

Consent

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Ethical approval

I declare on my honor that the ethical approval has been exempted by my establishment.

Sources of funding

None.

Author contribution

Ouafidi Btissam: Corresponding author, writing the paper and operating surgeon.

Kiram Hamza: writing the paper and operating surgeon.

Benaguida Hicham: writing the paper and operating surgeon.

Lamrissi Amine: study concept.

Fichtali Karima: correction of the paper.

Bouhya Said: correction of the paper and operating surgeon.

Guarantor

Dr. Ouafidi Btissam.

Registration of research studies

None.

Declaration of competing interest

The authors declare having no conflicts of interest for this article.

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