

SPONTANEOUS HETEROTOPIC PREGNANCY: A CASE REPORT

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ABSTRACT

Background: The simultaneous existence of an ectopic pregnancy and an intrauterine pregnancy is known as heterotopic pregnancy. These are identical twin pregnancies, where one nest is in the uterus and the other, wherever it may be, is ectopic. Although it is a rare pathology, there has been a noticeable increase in its occurrence in recent years due to the recurrence of high genital infections and the progress of medical procedures to help in reproduction. We report a case of heterotopic pregnancy. **Case Présentation:** A 29-year-old pregnant woman presented at 13 weeks to the emergency department with mild vaginal bleeding, and abdominal pain. Transvaginal ultrasound revealed a nine-week-old tubal pregnancy with an intrauterine thirteen-week-old gestational sac confirming HP. A salpingectomy was performed and the extrauterine pregnancy was successfully removed with the preservation of the intrauterine embryo. The course of the intrauterine pregnancy was uneventful and the patient gave birth to a healthy boy via vaginal delivery. **Conclusion:** Spontaneous heterotopic pregnancy is very rare. Early pelvic ultrasound during the first trimester should be routine to obtain an accurate diagnosis and proper management of such cases.

KEYWORDS: Heterotopic pregnancy, Ectopic pregnancy, Tubal pregnancy, Management, Salpingectomy.

INTRODUCTION

Heterotopic pregnancy, also known as ditopic or combined pregnancy, is defined as the coexistence of an intrauterine pregnancy and an extrauterine pregnancy.^[1] It is a pathological form of dizygotic twin pregnancy in which one twin is implanted in the uterine cavity and the other is implanted outside the uterus, the most frequent ectopic pregnancies being located in the fallopian tubes.^[2] The first case was described by Duverney in 1708 during an autopsy.^[3]

The incidence of GH in spontaneous pregnancies is approximately 1 in 30,000.^[4] However, with the increasing use of artificial reproductive techniques, such as multiple embryo transfers during in vitro fertilization (IVF), the development of medically assisted reproduction techniques, in particular the use of ovulation inducers, and the rising prevalence of pelvic inflammatory disease and tubal lesions, the incidence of GH has been estimated at 1.5/1000.^[5]

Diagnosis of a heterotopic pregnancy is a challenge for medical practitioners, although ultrasound is widely used and diagnostic difficulties remain.^[1]

The main treatment consists in extracting the ectopic pregnancy and preserving the intrauterine pregnancy.^[5]

The prognosis for the intra-uterine pregnancy and the mother depends on the speed of diagnosis, which ideally should be made before rupture of the ectopic pregnancy.^[6]

We report a case of heterotopic pregnancy collected in the Department of Gynecology-Obstetrics Oncology and High-Risk Pregnancy at the Rabat Maternity Hospital.

By analyzing this observation and the data available in the literature, we will review the epidemiological and pathophysiological aspects, the different clinical pictures of this pathology, the contribution of ultrasound in approaching its diagnosis, the possible therapeutic management modalities and the evolutionary aspect of heterotopic pregnancies.

CASE PRESENTATION

A 29-year-old woman, gravida 1, para 0, was referred to the emergency department, presenting at 13 weeks of gestation and 4 days with pain in the lower abdomen and intermittent vaginal bleeding for 2 days. She had no risk factors. Her medical history was unremarkable, and she had never surgeries on tubes or ovaries, no use of a fertility drug or hormone, and no intrauterine contraceptive device. On admission, she was hemodynamically stable and conscious, the pulse rate

was 70/min, blood pressure was 110/60 mmHg and her temperature was normal. The pain was located in the lower abdomen, her laboratory tests were normal except the hemoglobin was 10 g/dl.

Pelvic ultrasound showed a gravid uterus with a single live intrauterine fetus containing an embryo with a biparietal diameter equal to 22.5 corresponding to 13 SA and 5J with positive cardiac activity associated with the presence of a left latero-uterine adnexal mass, ovoid in shape, well defined with regular tapers, heterogeneous with a thickened wall taking color Doppler, measuring 58/32 mm with a gestational sac containing a non-evolutive pregnancy with an embryo estimated to be 9 SA in favor of an arrested ampullary extra uterine pregnancy. There was also a small effusion in the cul de sac of Douglas (Figure 1,2). The diagnosis of GH was confirmed.

Surgical exploration revealed: a 200cc hemoperitoneum and a ruptured left ampullary pregnancy (Figure 3). The right adnexa were normal. The patient had a left salpingectomy (Figure 4).

Anatomopathological examination of the surgical specimen confirmed tubal pregnancy.

Post-operative management was simple, and the patient was treated with natural progesterone 600 mg/d, divided into three doses, for six weeks. Pregnancy monitoring revealed no anomalies, and the patient delivered vaginally at term.

DISCUSSION

The coexistence of intrauterine and extrauterine pregnancies is a rare clinical phenomenon known as spontaneous heterotopic pregnancy. Seventy percent of HPs are typically discovered in the fallopian tubes between weeks five and eight of pregnancy. The diagnosis becomes less prevalent with increasing gestational age; 20% of diagnoses happen between 9 and 11 weeks, and less than 10% beyond 11 weeks.^[7]

From the physiopathological aspect, many hypotheses have been put forward to explain this double fertilization: it may be a spontaneous heterotopic pregnancy outside any context of hyperstimulation in relation to simultaneous fertilization or deferred fertilization, either superfecundation or superfetation, or during assisted reproduction techniques like ovulation inducers and in vitro fertilization and embryo transfer.^[8-9]

There are many causes of ectopic implantation of one of the fertilized eggs, including^[10-11]: Tubal causes: common to all ectopic pregnancies, either mechanical (acquired or congenital) or functional. Ovular causes: common to all ectopic pregnancies, either mechanical (acquired or congenital) or functional. These causes include tubal damage from pelvic inflammatory disease, previous ectopic pregnancy, endometriosis, tubal

surgery.^[12] In the current case, the patient had no risk factors.

HP can cause abdominal pain, vaginal bleeding, and spotting, as well as an adnexal mass in the context of an enlarged gravid uterus, with or without peritoneal irritation.^[13] In this case, the patient was asymptomatic until 13 weeks of pregnancy, and she complained of pain and bleeding for two days prior to surgery. A pelvic ultrasound revealed a mass in the left annex.

The clinical symptomatology is often mistaken for a threatened or ongoing abortion, and the diagnosis is only suspected on appearance of signs of hemoperitoneum secondary to rupture of the ectopic pregnancy, whether or not associated with a potentially fatal state of maternal shock.^[14]

Transvaginal ultrasound plays a significant role in diagnosis. This technology is operator-dependent, and it is possible that the examiner, in concentrating on the discovery of an intrauterine pregnancy, may neglect a complete examination of the annexes.^[15] Adnexal examination should be carried out systematically after evidence of IUP, especially in patients with risk factors for EP. A B-hCG test was not performed.

There are three approaches to treating heterotopic pregnancy. First, certain cases can resolve spontaneously, but there are no precise standards or measures to identify whether people fall into this category.^[16] The second option is medical treatment. The products used must be non-toxic for the remaining intra-uterine pregnancy, if evolutive. The most frequently used products are hyperosmolar glucose serum or sodium chloride injected locally, using laparoscopy or transvaginal ultrasound. Medical treatment can only be used if the ectopic pregnancy is young, asymptomatic, unruptured and clinically stable. Intramuscular injection of methotrexate represents an alternative if the diagnosis is made at an early stage, with a non-progressive or non-viable intra-uterine pregnancy and a stable hemodynamic state.^[17]

Treating the ectopic pregnancy surgically is the third option. Although laparotomies were frequently performed in the past, laparoscopies are currently preferred because they heal more quickly.^[18] Laparoscopic management compared to laparotomy should be used for individuals who are hemodynamically stable.^[12] The present case was treated with a radical procedure (salpingectomy) and was managed with laparotomy; no problems arose during or following the procedure.

The evolution of heterotopic pregnancy depends on the stage at which the diagnosis is made, and on how early the patient is treated. When the diagnosis is made early, the outcome is generally favorable: 30-75% of intrauterine pregnancies progress to term after treatment

of the EP.^[17] More systematic and advanced medical management reduced maternal mortality to 0.98%.^[19]



Figure 1: Trans-abdominal ultrasound showing simultaneous intrauterine and extrauterine sac.



Figure 2: Ultrasound showing measurements of the adnexal mass.

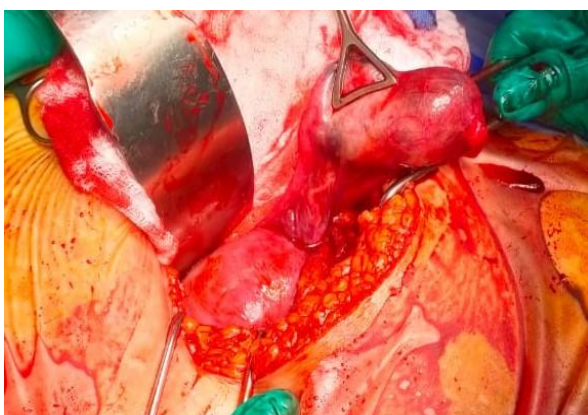


Figure 3: Peroperative image of tubal pregnancy with gravid uterus.



Figure 4: Exploration of the salpingectomy piece showing an amniotic sac with an embryo, a trophoblast and a hematosalpinx.

CONCLUSION

The diagnosis of heterotopic pregnancy should not be ruled out by the discovery of an intrauterine pregnancy during a spontaneous cycle. It should be suspected in the presence of abdominal pain, a latero-uterine mass with or without visible embryonic echo and intra-peritoneal effusion in the first trimester of pregnancy. Diagnosis must be made early to save the life of the patient and the intrauterine pregnancy. Ultrasound is the preferred para-clinical examination. The prognosis of intra-uterine pregnancy depends on early diagnosis and management.

RÉFÉRENCES

- 1 E. Albert Reece, M.D., Roy H. Petrie, M.D., Meredith F. Sirmans, M.D., Mieczyslaw Finster, M.D., and W. Duane Todd, M.D. Combined intrauterine and extrauterine gestations: A review (AM. J. OBSTET. GYNECOL, 1963; 146: 323.
- 2 Diallo D, Aubard Y, Piver P, Baudet Jh. Grossesse heterotopique : a propos de 5 cas et revue de littérature. J Gynecol obstet biol reprod, 2000; 29: 131-141.
- 3 Duverney G.J œuvres anatomiques (paris, jonbert), 1761 355,365.
- 4 Basile F, Di Cesare C, Quagliozzi L, Donati L, Bracaglia M, Caruso A, et al. Spontaneous heterotopic pregnancy, simultaneous ovarian, and intrauterine: a case report. Case Rep Obstetr Gynecol, 2012; 2012: 509694.
- 5 Barrenetxea G, Barinaga-Rementería L, Lopez de Larruzea A, Agirregoikoa JA, Mandiola M, Carbonero K. Heterotopic pregnancy: two cases and a comparative review. Fertil Steril, 2007; 87: 417 e419e415.
- 6 Laghzaoui Boukaidi M, Bouhya S, Sefrioui O, et all. Grossesses hétérotopiques : à propos de huit cas. Gynecol obstet fertil, 2002; 30: 218- 23.

- 7 I. Ramalho, I. Ferreira, J.P. Marques, M.J. Carvalho, A. Lobo, T. Rebelo, J. Paulo Moura, F. Aguas, Live birth after treatment of a spontaneous ovarian heterotopic pregnancy: a case report, *Case Rep. Womens Health*, Oct 10, 2019; 24: e00144. <https://doi.org/10.1016/j.crwh.2019.e00144>. PMID: 31709156; PMCID: PMC6833366.
- 8 CHOUKROUN J., FRIEDMEN S., CHENE P. Grossesse intra-uterine et extra-uterine simultanées. *J. Gynecol. Obstét. Biol. Reprod.*, 1981; 10: 231-34.
- 9 BOUSKINA M. Grossesse intra-uterine associée à une grossesse extra-uterine. A propos d'un cas. *These méd. Casablanca*, 1992; n° 254.
- 10 Kartikey. R. Shastri. KR, Anand. NI, Ganatra. BM. A rare case of heterotopic pregnancy in natural conception with ectopic pregnancy. *Indian Journal of Obstetrics and Gynecology Research*, 2015; 2: 174-175.
- 11 Nishat. F, Mubarak Al Badi. M, Rahman. M and al. Heterotopic pregnancy with natural conception; a rare event that is still being misdiagnosed: a case report. *Clinical Case Reports*, 2016; 4: 272–275.
- 12 L. Arsala, D. Danso, Spontaneous heterotopic triplet pregnancy with tubal rupture, *J. Invest. Med. High Impact Case Rep.*, 2014; 2(2), <https://doi.org/10.1177/2324709614531556>, 232470961453155
- 13 M. Aziz, J. Arronte, A case of spontaneous heterotopic pregnancy in natural conception complicated with hemoperitoneum, *Heliyon*, Feb. 10, 2020; 6(2): e03373. <https://doi.org/10.1016/j.heliyon.2020.e03373>. PMID: 32072059; PMCID: PMC7013160.
- 14 Simsek T, Dogan A, Simsek M, Pestereli E. Heterotopic triplet pregnancy (twin tubal) in a natural cycle with tubal rupture: a case report and review of the literature. *J Obst et Gynaecol Res.*, 2008; 34(4): 759-62.
- 15 Lilas Channiss a, Tala Tahle a, Rami Sabouni a,* , Mohammed Jamalih b, Heterotopic pregnancy with superfetation following ovarian stimulation: A case report. *Case Reports in Women's Health*, 2023; 40: e00562. journal homepage: www.elsevier.com/locate/crwh. <https://doi.org/10.1016/j.crwh.2023.e00562>.
- 16 M.D. Scheiber, M.I. Cedars, Successful non-surgical management of a heterotopic abdominal pregnancy following embryo transfer with cryopreserve-thawed embryos, *Hum. Reprod*, 1999; 14: 1375.
- 17 HANAFI. A; DAIF.L; LAMRISSIA; FICHTALIK;; BOUHYA.S Grossesses hétérotopiques : A propos de deux cas IJCRT, July 2021; 9(7). | ISSN: 2320-2882.
- 18 M. Michał, M. Marian, M. Marek, W.O. Ewa, Heterotopic pregnancy in the absence of risk factors—diagnostics difficulties, *Ginekol. Pol.*, Nov. 2011; 82(11): 866–868. PMID: 22384622.
- 19 E. Albert Reece, M.D., Roy H. Petrie, M.D., Meredith F. Sirmans, M.D., Mieczyslaw Finster, M.D., and W. Duane Todd, M.D. New York. Combined intrauterine and extrauterine gestations: A review. *AM. J. OBSTET. GYNECOL*, 1963; 146: 323.