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**Review Article**

# One Pregnancy Can Hide Another about a Case of Spontaneous Heterotopic Pregnancy Maternity, Ward of the Hospital Principal De Dakar

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## Abstract

**Introduction:** Heterotopic pregnancy, also known as ditopic pregnancy, is defined as the simultaneous occurrence of an intrauterine and extrauterine pregnancy. It occurs exceptionally during a natural cycle. Diagnosis is often late, at the stage of complications that can be life threatening. We report here the exceptional case of a spontaneous heterotopic pregnancy with a live ectopic embryo, diagnosed at an advanced stage by ultrasound. **Observation:** This was Mrs C.K.M 36 years old, 2nd gestity, 1st parity who was seen for pelvic pain, vomiting and 10 weeks of amenorrhea. The general condition was preserved with tenderness in the right iliac fossa without signs of peritoneal irritation. The emergency ultrasound scan showed a large haemoperitoneum and the coexistence of a progressive intrauterine mono-embryonic pregnancy of 10 weeks + 3 days and a right tubal ectopic pregnancy with a live embryo of 10 weeks + 5 days. A total right salpingectomy by laparotomy under tocolysis was performed urgently. The postoperative course was simple. The evolution was marked by a continuation of the intrauterine pregnancy without complications leading to a full term vaginal delivery of a live and healthy child.

**Conclusion:** Heterotopic pregnancy, a fairly frequent complication of medically assisted procreation, very rarely occurs during a spontaneous cycle. It should be suspected in the presence of any acute pelvic pain in the first trimester in an intrauterine pregnancy despite the absence of signs of peritoneal irritation or shock. Ultrasound in the first trimester of pregnancy remains the gold standard and allows the number of gestational sacs and embryos to be determined, but above all to ensure the intrauterine topography.

**Keywords:** Spontaneous pregnancy; Heterotopic; Pelvic pain; Ultrasound; Laparotomy

## Introduction

Heterotopic pregnancy, also known as ditopic pregnancy or combined pregnancy is defined as multiple gestation at two or more implantation sites [1]. It is a potentially fatal condition that rarely occurs during a natural conception cycle with an incidence of 1/30,000 versus 1% in medically assisted reproduction (MAP) [2]. Although exceptional, it has become more frequent since the development of medically assisted procreation (MAP), particularly ovulation inducers and the increase in genital infections and smoking. Diagnosis is often delayed, at the cost of life-threatening complications, which occur in the majority of cases before the 8th week of amenorrhea. Ultrasound and laparoscopy have revolutionized diagnosis. Management remains controversial [3].

The review of the literature only reported rare African publications, including one in Senegal in 2013 [3]. Thus, we report here the exceptional case of a spontaneous heterotopic pregnancy, which was diagnosed by the visualization of the simultaneous presence of two embryos with cardiac activity in the uterine cavity and in the right tubal ampulla.

## Observation

This was Mrs C.K.M, 36 years old, primiparous, who was seen for pelvic pain for 3 days associated with a few episodes of vomiting in a 10-week amenorrhea. It was a spontaneous conception and the patient had no history of abortion, infertility, pelvic inflammatory disease or abdominal surgery. Active and/or passive smoking was not found on examination. On examination, the general condition was preserved with a blood pressure of 12/7 cm Hg, a pulse of 92 bpm, tenderness in the right iliac fossa with no signs of peritoneal irritation. On vaginal touch, the uterus was enlarged with perception of a right latero-uterine mass. The fingernail came back clean. The emergency ultrasound scan showed a large haemoperitoneum and the coexistence of a progressive intrauterine mono-embryonic pregnancy of 10 weeks + 3 days and a right tubal ectopic pregnancy with a live embryo of 10 weeks + 5 days (Figures 1-3).

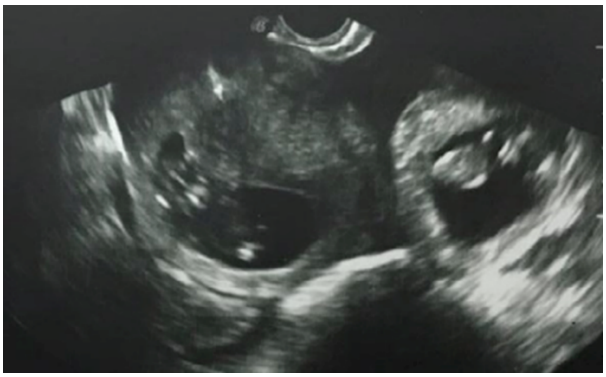

**Figure 1:** Ultrasound image.

The blood count showed a haemoglobin level of 8.5g/dl. Emergency laparotomy under tocolysis revealed a large haemoperitoneum estimated at 650 ml, a fissured right ampullary tubal pregnancy with live embryo, a gravid uterus and persistent corpora lutea on both ovaries. A total right salpingectomy was performed.

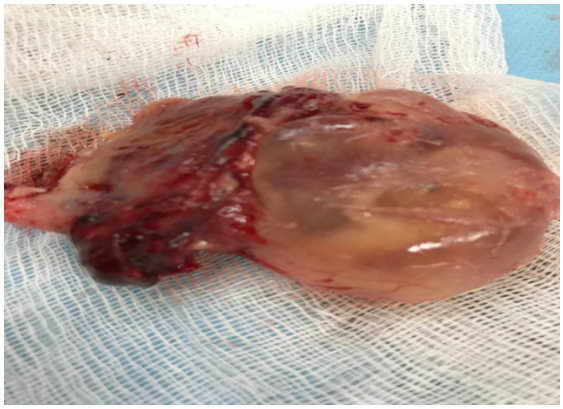

**Figure 2:** Surgical specimen.

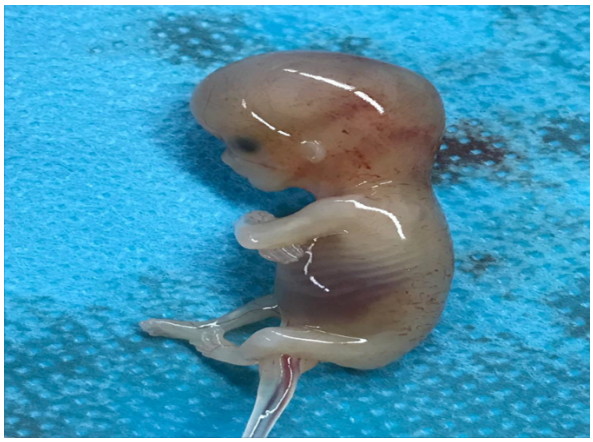

**Figure 3:** Ectopic embryo.

The postoperative course was straightforward with the patient discharged at D4. The surgical specimen was sent to anatomopathology, which confirmed the diagnosis. The short and medium term evolution was marked by a continuation of the intrauterine pregnancy without any symptoms or complications. The pregnancy was well monitored with four unremarkable antenatal visits. The patient gave birth at term (39SA) by vaginal delivery of a healthy newborn weighing 3180g, without complications.

## Discussion

The first case of heterotopic pregnancy was reported in France by Duverney in 1708 following an autopsy [2,4]. The second description was made in 1761 during the autopsy of a woman in the third month of pregnancy and the third a century later [5,6]. Heterotopic pregnancy is defined as a multiple gestation at two or more implantation sites. It can include two ectopic pregnancies, but most often one of the pregnancies is intrauterine [1]. However, the most common definition of heterotopic pregnancy is the coexistence of one or more intrauterine pregnancies (IUPs) with an ectopic pregnancy (EP), regardless of its location [7]. The intrauterine pregnancy is most often unique, nevertheless a case of heterotopic pregnancy with an ectopic (abdominal) pregnancy associated with a multiple intrauterine pregnancy was described by Aguemon and al. in Benin in 2015 [5], Ben Temime and al. in Tunisia in 2012 [8] and Bataille and al. in France in 2017 [9]. Ectopic pregnancy can be tubal, ovarian, cervical, cornu or abdominal. Approximately 1% of pregnancies occur in an ectopic location, 95-97% of which are in the fallopian tube. The most common site is the ampulla of the tube (80%), followed by the isthmic segment of the tube (10%), the tubal fringes (5%) and the cornual and interstitial regions (2%-4%) [2]. The majority of heterotopic pregnancies described in the literature had an ampullary location as was the case with our patient. However, other locations of EP in the context of heterotopic

pregnancy have been published: abdominal (Aguemon and al. [5] and Sanogo and al. [6]); ovarian (Basile and al. [10], Radhouane and al. [11] and Laghzaoui and al. [12]); isthmic (Traoré and al. [13]) and corneal (Chadee and al. [14]). Heterotopic pregnancy is a rare but not exceptional condition with an estimated frequency of between one in 20,000 and one in 30,000 in natural conception [2]. Its frequency is tending to increase with the development and widespread use of MAP techniques. These techniques are in themselves a source of EP with an increase in incidence to 1% [2]. The real frequency is probably underestimated. Indeed, heterotopic pregnancy may go unnoticed in the context of an EP associated with a spontaneous abortioń or that of an evolving UGI combined with a spontaneously resolved EP [15]. In Senegal, the frequency of heterotopic pregnancy has decreased from 1 in 2300 deliveries in 1994 to 1 in 4168 deliveries in 2009, thanks to the early and adapted diagnosis and treatment of upper genital infections [4]. The actual etiology of heterotopic pregnancy is still unknown [3]. Several theories have been put forward to explain the occurrence of this particular form of pregnancy. Pathophysiologically, combined pregnancy may result from simultaneous fertilisation (difference in the speed of migration of two fertilised eggs) or delayed fertilisation (fertilisation of two eggs produced at a short interval during the same cycle by two spermatozoa from two successive coitus. The risk factors for heterotopic pregnancy are those for EP. Common factors that predispose to the occurrence of an ectopic pregnancy are: smoking, endometriosis, progestin-only contraception, the morning-after pill, tubal surgery and pelvic inflammatory disease [7,15]. Genital infection is the main risk factor, especially subacute or chronic infections that go unnoticed, where Chlamydia plays an important role]. In addition, contraception with an IUD appears to be particularly associated with ovarian pregnancies [2,7]. EP is common in pauciparous women [7] as was the case with our patient. A history of EP is an important risk factor, particularly in the case of medical treatment of EP with methotrexate [2]. The evolution of MAP techniques has largely modified the epidemiological profile of heterotopic pregnancies [2,7,15,16]. Numerous studies have shown that pelvic inflammatory disease, previous tubal surgery, ovarian stimulation and MAP confer a higher risk of EP and hence heterotopic pregnancy [6]. In the majority (71%) of cases, risk factors for heterotopic pregnancy were found [17]. In our patient, no risk factors were found.

Heterotopic pregnancy, an often unrecognised condition, poses a diagnostic problem and can be life threatening for the woman [6]. The circumstances of discovery are variable. The diagnosis is easy when the signs of EP are in the foreground, the clinical symptomatology is then dominated by the classic triad of EP: amenorrhoea, metrorrhagia in 50% of cases, pelvic pain in 82.7 to 90% of cases. This was the case with our patient. Up to 33% of patients may initially present with haemodynamic instability [1]. The association of this triad with an increase in uterine volume is strongly suggestive of heterotopic pregnancy. The diagnosis is more difficult if the clinical picture is that of an intrauterine pregnancy. The clinical symptomatology is often associated with a threatened abortion or an ongoing abortion, and the diagnosis of heterotopic pregnancy is not made until signs of haemoperitoneum secondary to rupture of the EP, which may or may not be associated with maternal shock, which is often fatal. Finally, heterotopic pregnancy can be totally asymptomatic, discovered by chance during an ultrasound examination or intraoperatively [1,11,16]. The preoperative diagnosis of heterotopic pregnancy remains a major challenge for modern reproductive medicine. Although signs and symptoms such as abdominal pain, adnexal mass, peritoneal irritation, metrorrhagia and increased uterine volume have been reported to be predictive of heterotopic pregnancy, they are not specific and may be confused with other normal or abnormal manifestations of pregnancy. Furthermore, in the absence of MAP, the suspicion of multiple or heterotopic pregnancy is usually very low. Pelvic ultrasound, a simple and accessible examination, allows the diagnosis to be made. Visualisation of intrauterine and extrauterine cardiac activity, as in our patient and that of Hyun-Soo Jeon and al. [2], confirms the diagnosis of heterotopic pregnancy. Clinical signs of rupture (shock and peritoneal irritation), as well as haemoperitoneum, may facilitate the diagnosis as described by Muhammad Aziz and al. [18].

Seventy percent of heterotopic pregnancies are diagnosed between 5 and 8 weeks of gestation, 20% between 9 and 11 weeks, and less than 10% after 11 weeks [1,9]. Indeed, diagnosis was early for Nishat Fatema and al. (06 weeks+5D); Angela J. Chan and al. (07weeks); Basile and al. (07weeks); Migle and al. (07weeks); Radhouane and al. (08weeks+4D); Traoré and al. (08weeks+5D); Ouafidi and al. (09weeks+2D) [1,10,11,13,19-21]. In our patient, the diagnosis was made at 10 weeks of gestation as in Vanita and al. [22] and Veli Mihmanli and al. [23]. Heterotopic pregnancy can be unrecognised, particularly in abdominal forms with no or late diagnosis, as was the case described in Mali by Sanogo and al. where the abdominal pregnancy was discovered three months after the birth of the twin brother [6]. Endovaginal ultrasound allows the diagnosis of heterotopic pregnancy in 89% of cases [11]. However, it can give false assurance (33%) [17]. in the face of minor clinical signs of EP by just showing a normal intrauterine pregnancy, due to its variable sensitivity (26.3% to 92.4%) and operator dependency [3]. This highlights the complexity of the diagnosis. When ultrasound misses the diagnosis, only surgery can correct the diagnosis. The study by Talbot et al. showed that a surgical approach, either laparoscopy or laparotomy, was the cause of the diagnosis in 11% and 18% of cases respectively [17].

Heterotopic pregnancy carries a considerable risk of maternal morbidity and mortality due to the risk of rupture of the ectopic pregnancy. After a diagnostic challenge, the therapeutic challenge

remains to eliminate the EP while preserving the UGI. Surgery is the standard treatment, but non-surgical approaches are increasingly common. According to the literature, therapeutic modalities for heterotopic pregnancy include, in addition to surgery, abstention, methotrexate for patients who do not wish to preserve the UGI and injection of potassium chloride or hyperosmolar glucose or methotrexate into the ectopic sac and ultrasound-guided aspiration of the ectopic embryo [1,14]. However, due to the rarity of the condition, with most publications being case reports or small case series, there is no consensus on management [3], although conservative surgery preferably laparoscopic is considered the gold standard [11]. It is important to analyse the surgical specimen, which in exceptional cases may be a hydatidiform mole as described by Vanita and al. (partial mole) [22], with great impact on management. The complications of treatment are haemorrhagic and infectious [4]. The main concern remains the fate of UGI. When treated correctly and in time, a heterotopic pregnancy can result in a live birth with a favourable outcome for the child and the mother [20]. Indeed, 30-75% of UGI pregnancies progress favourably to term delivery [4,15] as was the case with our patient, Tingi and al. [24], Migle and al. [20], Kumar and al. [25], and Ben Temime and al. [11]. Preterm delivery may also occur as reported by Niang and al. [4] and Talbot and al. [17]. Treatment of EP may be complicated by abortion of the UGI. In the series by Talbot and al. 26% of pregnancies ended in miscarriage [17]. The same is true for Veli Mihmanli and al. (pregnancy terminated at 18 AU after salpingectomy via laparotomy) [23], Niang and al. (two spontaneous miscarriages after salpingectomy via laparotomy [4] and Muhammad Aziz and al. (abortion secondary to partial salpingectomy via laparoscopy) [18]. It is noteworthy that a higher rate of abortion of intrauterine pregnancy has been documented in patients undergoing medical versus surgical treatment, ranging from 50% to 13%. [18]. The route of delivery is most often vaginal. According to the literature, the caesarean section rate is 20% in relation to surgical treatment of cone-shaped EPs [14,15].

## Conclusion

Heterotopic pregnancy, a fairly frequent complication of medical procreation, very rarely occurs during a natural cycle. It must be suspected in the presence of any acute pelvic pain in the first trimester in an intra uterine pregnancy despite the absence of signs of peritoneal irritation or shock. Ultrasound in the first trimester of pregnancy remains the gold standard and allows the number of gestational sacs and embryos to be determined, but above all to ascertain the intrauterine topography. Because of its significant morbidity and mortality, it is crucial to recognize a heterotopic pregnancy at an early stage so that management, which is certainly not consensual, can be carried out without delay.

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