

# Ectopic Molar Pregnancy with Coexistent Normal Intrauterine Pregnancy: A Report of an Unusual Diagnosis managed in Yaoundé Gyneco-Obstetric and Pediatric Hospital, Cameroon

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## Abstract

**Background:** Heterotopic pregnancy is a very rare event in a natural cycle. Its incidence is rising mostly due to induction of ovulation or Assisted Reproductive Technique. Most ectopic in heterotopic pregnancies are localized in the fallopian tubes. The occurrence of molar tubal ectopic pregnancy coexisting with intrauterine pregnancy is uncommon and consequently not often considered a diagnostic possibility. **Case Report:** We report the case of a 25-year-old woman, gravida 4 para 1, who complained of vaginal spotting and lower abdominal pain after 6 weeks of amenorrhea following clomiphene citrate ovarian stimulation. Transvaginal Ultrasonography revealed an intrauterine pregnancy and an unruptured left tubal ectopic pregnancy. She underwent successful laparoscopic salpingectomy while the intra-uterine pregnancy was allowed to continue. The ectopic pregnancy tissue histology reported an ectopic partial molar pregnancy. She subsequently had a normal vaginal delivery at 39 weeks and 3 days and her follow-up was uneventful. **Conclusion:** Heterotopic pregnancy is a rare but life-threatening condition. It should be suspected in a pregnant woman with a risk factor of multiple gestations who presents vaginal bleeding and lower abdominal pain in the context of early pregnancy. Laparoscopic surgery is effective for confirming

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the diagnosis and treating the ectopic component. Routine histological examination of tubal specimens must be taken very seriously because some findings like molar pregnancies may require specific management.

## Keywords

Ectopic Molar Pregnancy, Heterotopic Pregnancy, Laparoscopy Surgery, YGOPH

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## 1. Introduction

Heterotopic pregnancy is a combination of intrauterine pregnancy and ectopic pregnancy. Its occurrence in a natural cycle is extremely rare with an incidence estimated to be approximately 1/30,000 pregnancies [1]. However, this incidence is tending to increase with Assisted Reproductive Technology (ART) between 1/100 and 1/500 [2]. Although the cause of heterotopic pregnancy varies from one woman to another, some of the most common risk factors are a history of Pelvic Inflammatory Disease (PID), surgical treatment for infertility, and forces generated during embryo transfer in the uterus. The frequency of hydatidiform moles is 1% - 2% of all pregnancies [3]. An ectopic pregnancy occurs in 1/1000 to 1/500 pregnancies [4]. The combined incidence therefore of molar ectopic pregnancy should be somewhere between 1 in 10,000 and 1 in 20,000 pregnancies [5]. Thus, the occurrence of molar tubal ectopic pregnancy coexisting with intrauterine pregnancy is consequently a rare situation. There are few reported cases of ectopic molar pregnancy combined with normal intrauterine pregnancy in the literature. With the patient's consent, we present a case managed in our hospital.

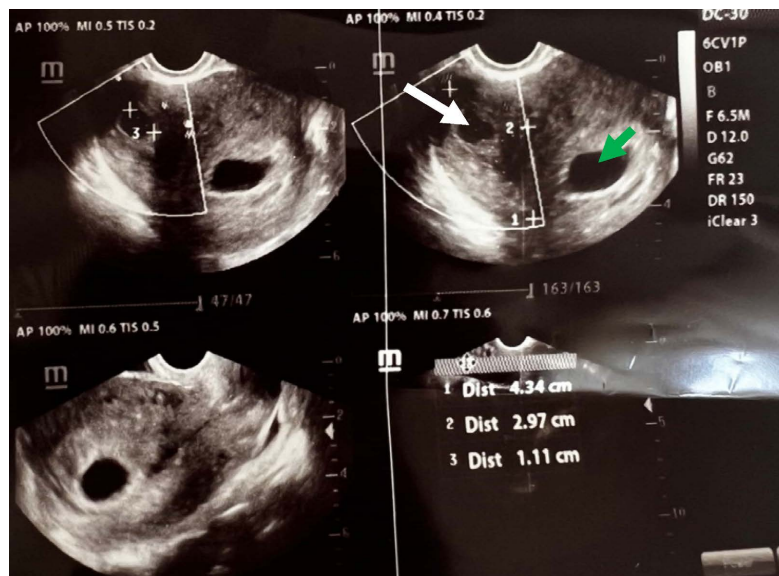
## 2. Case Presentation

The patient was a 26-year-old Cameroonian woman, Gravida 4 Para 1 with 2 previous uncomplicated induced abortions and 1 term pregnancy and live birth 5 years earlier. The patient was born a twin herself and has a family history of multiple pregnancies. She had a 2-year history of secondary infertility. Investigations of this infertility revealed a recurrent chlamydial infection and ovulatory disorder. Clomiphene citrate (100 mg/day for 5 days) had been prescribed for 3 months before conception. The rest of her past history was unremarkable. She presented to the Yaounde Gyneco-Obstetric and Pediatric Hospital (YGOPH)—Cameroon, with vaginal bleeding, lower abdominal pain, and 6 weeks of amenorrhea corresponding to her last menstrual period. The urinary pregnancy test was positive. On examination, her pulse was 87 beats/min and regular; her blood pressure was 105/70 mmHg. Abdominal examination revealed no distension but mild tenderness at the right lower abdomen. A pelvic examination revealed a spotting through the cervix. Cervical motion tenderness was present. The uterus was slightly enlarged. Both right and left adnexa were difficult to ap-

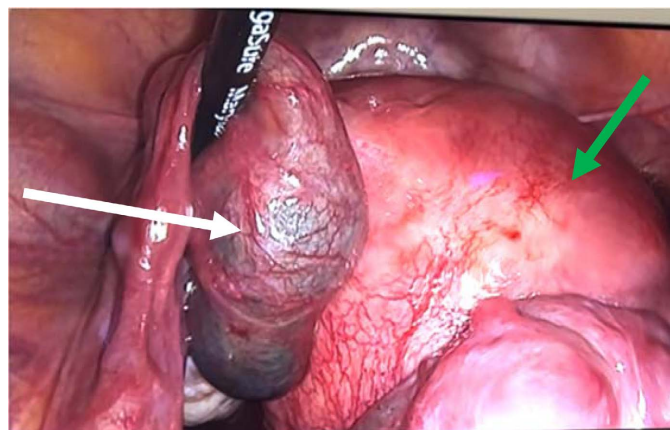
preciate because of tenderness. An assessment of ruptured ectopic pregnancy was made.

Laboratory investigations indicated anemia, with hemoglobin of 9.2 g/dl and hematocrit of 29.3%. Serum  $\beta$ -hCG was not performed. Transvaginal ultrasonography (**Figure 1**) showed an intrauterine pregnancy and left adnexal mass with the presence of free fluid in the Douglas pouch.

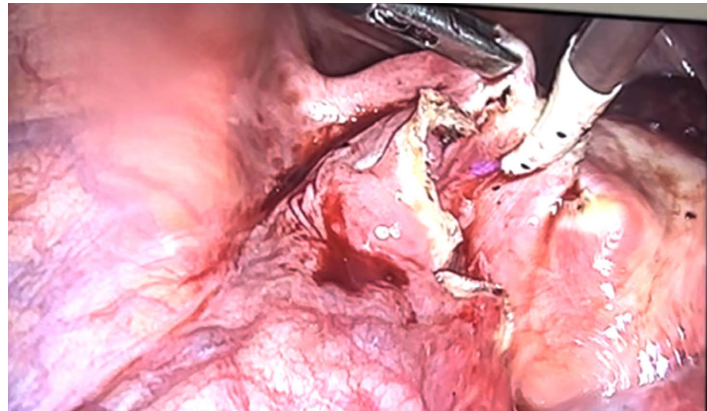
A diagnosis of heterotopic pregnancy with a ruptured ectopic component was made and laparoscopic surgery with low pressure (10 mmHg) and without uterine cannulation was performed. A Hemoperitoneum of about 100 ml was found. The uterus was consistent with 6 weeks of gestational age and the right adnexa was normal. The left fallopian tube was not ruptured but was markedly distended through the junction between the isthmus and ampulla with tubal abortion (**Figure 2**). Left total salpingectomy was carried out (**Figure 3**).



**Figure 1.** Pelvic ultrasound image showed heterotopic pregnancy Intrauterine pregnancy (green arrow) and tubal pregnancy (white arrow).



**Figure 2.** Laparoscopic view of the left tubal pregnancy (white arrow); gravid uterus (green arrow).



**Figure 3.** Laparoscopic view of the left salpingectomy.

The systematic histological examination showed chorionic villi of variable size on about 70% of the fields, most of them were moderately hydropic and map-shaped. They all present an epithelial coating with moderate hyperplasia, somewhat irregular but without cytonuclear atypia. The few vessels found are sometimes dilated by red blood cells and create meandering images. Besides these histological elements, some tubal flaps are found with cells without atypia. All this was consistent with ectopic partial molar pregnancy.

Unfortunately, we don't have the technology to provide a microscopic picture of the histology. Serum  $\beta$ -hCG level was not asked for follow-up because of viable intrauterine pregnancy. Chromosomal karyotyping and molecular cytogenetic analysis of chorionic villi were not performed because of cost issues and the availability of these exams. After two days, the patient was discharged, and we allowed the intrauterine pregnancy to continue. During the postoperative period, subsequent clinical and ultra-sonographic courses were normal. She had a normal vaginal delivery of a healthy male infant at 39 weeks 3 days and her follow-up was uneventful with normal  $\beta$ -hCG level at the late postpartum period, three and six months after delivery.

### 3. Discussion

Normal intrauterine pregnancy with a coexisting molar ectopic pregnancy is extremely rare. Yet, heterotopic pregnancy is likely to more frequently occur with the increasing number of multiple births due to couples undergoing treatment for fertility [6]. Most heterotopic pregnancies occur as a result of Assisted Reproductive technology (ART). There are many risk factors for heterotopic pregnancy, such as previous ectopic pregnancy, cigarette smoking, in vitro fertilization (IVF), gamete intrafallopian transfer (GIFT), ovulation induction, and tubal damage after Pelvic Inflammatory Disease, endometriosis or previous tubal surgery [7] [8]. With these risk factors, heterotopic pregnancy should be kept in mind and investigated. Also, a few heterotopic pregnancy cases have been reported in natural cycles [9] [10]. In our case, the patient's risk factors include a family history of multiple pregnancies, Which increases her risk of multiple

pregnancies, being a twin herself, and using clomiphene citrate to induce ovulation, which also increases the risk of multiple pregnancies ovulations and multi-fetal pregnancy as this patient had. The location of a fertilized egg outside the uterus can vary, but it tends to end up in one of the fallopian tubes in 72.5% of cases [11]. An important risk factor for ectopic pregnancy in this patient was her history of recurrent chlamydial infection. Recurrent Chlamydial infection may be associated with potential tubal damage especially when not properly treated and a significant risk factor for ectopic pregnancy [12]. This may partly be responsible for the ectopic pregnancy this patient had.

The combined incidence of molar ectopic pregnancy should be somewhere between 1 in 10,000 and 1 in 20,000 pregnancies [5]. A diet low in vitamin A and in animal fat, previous molar pregnancy, maternal A or AB blood group while father O group, age > 40 years, and a history of recurrent spontaneous abortion appear to be associated with an increased risk of developing molar pregnancy [13]. Our patient was from "A" blood group while her husband was from the O blood group. Tubal molar pregnancy is clinically indistinguishable from a traditional tubal pregnancy, but with a higher tendency of rupture at the time of presentation, making the diagnosis difficult [14].

Symptoms and signs in our case were lower abdominal pain, vaginal bleeding, adnexal mass, and an enlarged uterus. These clinical features are not different from those seen in traditional extra-uterine pregnancy although Tal *et al.* [2] noted that there was no vaginal bleeding in fifty percent of cases; probably because of the existing intrauterine pregnancy. High-resolution trans-vaginal ultrasound is the most important aid in the diagnosis of heterotopic pregnancy. Previous reports [2] noted that 70% of cases were diagnosed between 5 and 8 weeks of gestation, 20% between 9 and 11 weeks, and 10% after the 11<sup>th</sup> week of gestation. The current case was at 6 weeks of gestation. Ultrasound identifies less than 50% of hydatidiform moles, with a detection rate higher for complete compared to partial moles, and improves even further after the 14<sup>th</sup> week of gestation [15]. Although histopathological examination of the conceptus remains the current gold standard for the diagnosis of gestational trophoblastic diseases, no single diagnostic method can confirm the presence of a mole with 100% accuracy. A tubal molar pregnancy can be over-diagnosed by histological examination, because it is difficult to differentiate from non-molar hydropic abortions and early placentation, as non-molar tubal pregnancies may also exhibit hydropic villi [4]. Therefore, to reduce the incidence of this over-diagnosis, a combination of histological features and DNA flow cytometry analysis is necessary for suspected tubal ectopic hydatidiform mole [4].  $\beta$ -hCG alone is not helpful in the diagnosis of heterotopic pregnancy. Although  $\beta$ -hCG levels are elevated in tubal molar pregnancies, they are generally in the lower range, because implantation in the fallopian tube might preclude adequate vascularization, thereby leading to low levels of  $\beta$ -hCG. There is no distinctive difference in  $\beta$ -hCG levels between molar tubal pregnancies and ectopic pregnancies. Thus, an early ectopic

molar pregnancy is not distinguishable from a non-trophoblastic tubal pregnancy based on  $\beta$ -hCG levels [16]. For this reason,  $\beta$ -hCG was not done in our case for the follow-up of the patient postoperatively. We believe that with an ongoing viable intrauterine pregnancy, follow-up with  $\beta$ -hCG will be not helpful. In our case,  $\beta$ -hCG was negative in the late postpartum period, three and six months after delivery. This allowed us to assess the absence of persistent proliferative activity of trophoblast.

The management of heterotopic pregnancy remains controversial. The ectopic component is usually treated surgically, whereas the intrauterine component is expected to develop normally. The most important issue in surgical treatment in heterotopic pregnancy is the safe removal of the extra-uterine pregnancy and the maintenance of the normally located intra-uterine pregnancy. The laparoscopic approach is preferred over laparotomy because of its several advantages, which include less bleeding, fewer surgical wounds, less pain, reduced requirement for antibiotics and analgesics, short hospitalization, and, quicker recovery time [17]. However, it must be underlined that special circumstances such as a large uterine size, and physiological changes in the pregnant woman may make laparoscopic surgery more difficult and increases the risk for complications. Our patient was only 6 weeks pregnant which made laparoscopy the preferred route of treatment. For the tubal component of heterotopic pregnancy, salpingectomy is indicated over salpingostomy [17] because the latter may result in incomplete removal and persistent ectopic pregnancy, which will be impossible to identify with serial  $\beta$ -hCG levels given the existing intra-uterine pregnancy. Because most heterotopic pregnancies are found during the first trimester, controversy exists regarding the maternal and fetal effects of anesthetic agents and pneumoperitoneum [18]. However, according to the Society of American Gastrointestinal Surgeons, the effects on pregnant women and fetuses are small if the operation time is less than 60 minutes and the intraperitoneal pressure is maintained below 10 - 12 mmHg [18] [19]. Recently, studies have shown that gasless laparoscopy has proven to be a safe alternative to conventional laparoscopy for gravid patients [20] [21]. In this case, the patient underwent left laparoscopic salpingectomy with low (10 mmHg) intraperitoneal pressure; the operative time was 35 minutes and fortunately, the postoperative period was uneventful. With early diagnosis and treatment, 70% of intrauterine pregnancies will reach viability [22].

#### 4. Conclusion

Heterotopic pregnancy is a rare life-threatening event. It should be suspected in women with risk factors of multiple gestations who present signs and symptoms of ectopic pregnancy. Laparoscopic surgery performed by an experienced surgeon is a feasible and safe surgical modality for the treatment of the ectopic part of the heterotopic pregnancy. It is important and recommended to conduct a systematic histological examination of tubal specimens in ectopic pregnancy to diag-

nose cases of ectopic molar gestations early and initiate appropriate post-treatment surveillance.

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## Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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