# Case Report of an Ectopic Molar Pregnancy in the Presence of an Intrauterine Device

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# **ABSTRACT**

**Background:** Ectopic molar pregnancy is a rare phenomenon and has not been reported in the presence of an intrauterine device (IUD). Clinical diagnosis of molar pregnancy is challenging and requires careful follow-up.

**Case:** A 25-year-old woman (gravida 2, para 0) with a copper IUD in place presented with a positive pregnancy test. Diagnosis of the complete hydatidiform mole was pathologically confirmed after surgery following clinical and sonographic investigations that identified a left-sided ectopic pregnancy.

**Conclusion:** Gestational trophoblastic disease (GTD) presenting as an ectopic pregnancy is a very rare occurrence. This patient recovered without event through a combined management and follow-up for ectopic pregnancy and gestational trophoblastic disease. Appropriate identification and management of this clinical problem is essential in order to prevent initial complications as well as subsequent malignant sequelae.

## INTRODUCTION

Ectopic gestation is a fairly common phenomenon, whereas ectopic molar pregnancy is extremely rare, affecting between 1 in 10,000 and 1 in 200,000 pregnancies. Main risk factors for molar pregnancy include a history of gestational trophoblastic disease (GTD) and increasing age. Other risk factors include a history of spontaneous abortion, long-term use of oral contraceptives, being of Asian or American Indian descent, or being African American. Complete moles occur when an empty ovum is fertilized by a single sperm that undergoes complete reduplication

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of the haploid genome immediately after entering the egg, or it can occur when an empty ovum is fertilized by 2 sperm.4 This most commonly results in a 46 XX trophoblast.<sup>5</sup> Trophoblastic proliferation is associated with diffuse villous edema and the absence of fetal tissue. A wide range of nonmalignant complications in a patient is possible including anemia, infection, hyperthyroidism, pregnancy-induced hypertension, and coagulopathy, and less frequently, pulmonary complications.6 Some variation of these associated issues is seen in approximately 25% of patients diagnosed with GTD. Malignant sequelae is seen 20% of the time following evacuation of a complete molar pregnancy. The

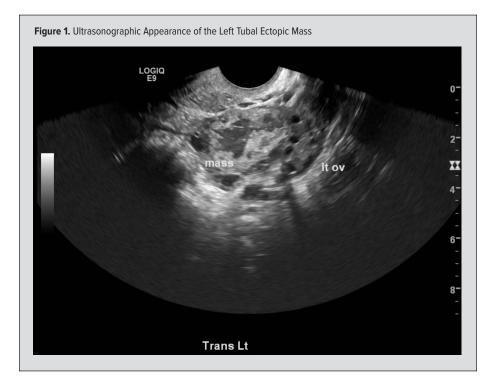
likelihood of malignant outcomes for a patient can be determined by use of a prognostic scoring system.<sup>5</sup>

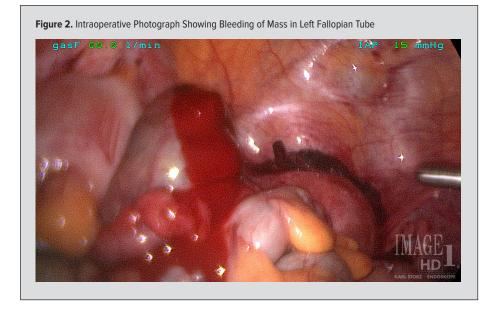
Risk factors for ectopic pregnancy include a history of ectopic pregnancy, previous tubal surgery, in utero diethylstilbestrol (DES) exposure, and documented tubal abnormalities. Less important risk factors are a history of pelvic infections, a history of infertility, and more than 1 lifetime sexual partner.<sup>7</sup>

Clinical diagnosis of molar pregnancy is challenging and judicious follow-up monitoring is required. Herein we describe the first case known to us of an ectopic molar pregnancy with a copper IUD in place.

### **CASE PRESENTATION**

A 25-year-old woman (para 2, gravida 0) presented to her local family planning clinic 6 weeks after her last menstrual period with a positive pregnancy test. She had a copper IUD in place at the time of presentation, which was subsequently removed. A few days later, she had an ultrasound that demonstrated no sonographic evidence of an intrauterine or ectopic pregnancy.





However, she was contacted later in the day with her quantitative  $\beta$ -human chorionic gonadotropin (hCG) results of 40,000 U/L and was instructed to present to her local emergency department. Once there, she was advised she most likely had had a miscarriage and she was instructed to follow up in obstetrics/gynecology clinic. At that time, she denied any pain and her vaginal bleeding was minimal.

A pelvic ultrasound (Figure 1) was obtained 2 days later at her scheduled clinic follow-up. The sonographic findings of a left-sided ovarian mass were consistent with a left-sided ectopic pregnancy. Laboratory investigations indicated a quantitative  $\beta$ hCG of 31,356 U/L. She was sent to the hospital immediately

for surgical management of presumed ectopic pregnancy.

Laparoscopy revealed a small amount of blood in her pelvis and dilation of the ampulla of the left fallopian tube. With gentle examination of the tube, profuse bleeding occurred (Figure 2), and the decision was made by the obstetriciangynecologist to perform a salpingectomy. Subsequent pathology findings revealed a ruptured ectopic pregnancy with complete hydatidiform mole.

Gynecologic Oncology at the regional university hospital was consulted. They recommended suction dilation and curettage (D&C) to ensure no molar tissue was found in the uterus. The patient underwent an uncomplicated D&C the following week, and pathology was negative for molar tissue. Follow-up included weekly blood draws to track quantitative BhCG values down to zero, after which time monthly blood draws were completed to ensure BhCG values remained negative for 6 months. She was instructed about the importance of reliable contraception during the follow-up period, and was given a prescription for oral contraceptive pills.

# **DISCUSSION**

Ectopic GTD is an exceptionally rare occurrence. The presence of a coppercontaining IUD with a molar pregnancy makes this case even more exceptional, and to our knowledge, this has not been reported previously in the literature. Only 0.8% of women experience pregnancy in the first year of use of a copper IUD, making it one of the most effective forms of

contraception available.<sup>8</sup> The risk of ectopic pregnancy is higher in women who become pregnant with an IUD in place compared with women who do not have an IUD. However, ectopic pregnancies occur less often in IUD users than in women using other methods of contraception or not using any contraception, because IUDs are highly effective at preventing pregnancy.<sup>9</sup> In a previous prospective study, Honore' suggested a selective suppression of molar pregnancy development by the presence of an IUD.<sup>10</sup> The researchers examined pathology from all spontaneous abortions during the study period and found there were no molar pregnancies in the group that had IUDs in place.

It is proposed that the management and prognosis of ectopic

GTD is similar to that of ectopic pregnancy and GTD combined. There is some literature to suggest that ectopic GTD has a higher likelihood of rupture at the time of presentation versus nontrophoblastic ectopic pregnancy.<sup>11</sup> If there is suspicion for molar ectopic pregnancy preoperatively, salpingectomy should be considered over salpingostomy to ensure removal of all of the molar tissue. Use of a laparoscopic bagging device to remove the ectopic pregnancy from the patient would be beneficial to prevent spillage into the abdomen.

This case demonstrates successful management and follow-up of ectopic GTD without recurrence of disease or malignant conversion by combining the recommendations for the management of ectopic pregnancy and gestational trophoblastic disease. The confluence of extremely rare circumstances surrounding this molar tubal ectopic pregnancy in the presence of a highly effective contraceptive device is remarkable and virtually unknown in the medical literature. Though ectopic GTD is extremely rare, the possible complications from missing this diagnosis and having inadequate treatment and follow-up highlight the need for conclusive pathologic investigation of all removed gestational tissue, whether intra- or extra-uterine.

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