

# CASE REPORT: HETEROTOPIC PREGNANCY IN A 12-YEAR-OLD ADOLESCENT

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#### **ABSTRACT**

Background: Heterotopic pregnancy is a rare entity in which it is defined by a topical (intrauterine) pregnancy concomitant with an ectopic pregnancy (outside the uterus, usually in fallopian tubes). While hydatidiform mole is characterized by a benign tumor that develops in the uterus as a result of a non-viable pregnancy. The diagnosis is made by means of β-HCG measurements and ultrasound examination. Therapeutic management is based on the viability of the topical pregnancy and the patient's conditions. OBJECTIVE: To emphasize the importance of diagnosing heterotopic pregnancy, to decide the best management that includes therapeutic success, fertility preservation, and maintenance of intrauterine pregnancy. Case Report: A 12-year-old patient with no history of morbidity, primiparous, smoker, complaining of mild lower abdominal pain, especially in the right iliac fossa, and denying transvaginal bleeding, had transvaginal ultrasonography (US) showing a 12-mm left attached image with fluid and 7-mm yolk bladder. The patient had beta-hCG > 15,000 mIU/mL and an adnexal mass of 12 mm with no intrauterine alterations, and exploratory laparotomy was performed, at which time the presence of a moderate amount of blood in the abdominal cavity was identified, but normal fallopian tubes and ovaries bilaterally, and no gestational sac in the abdominal inventory. On the first postoperative day, the patient developed diffuse abdominal pain of moderate intensity without peritonism. On the second postoperative day, the patient evolved with significant improvement in abdominal pain and bowel movements. With the result of the new Beta-HCG (>15,000 mIU/mL), presenting the same value as before, a new transvaginal ultrasound is requested: uterine cavity with echogenic endometrium, thickened of 27 mm, irregular and with anechoic areas intermingled. Therefore, gestational trophoblastic disease/Ovular remains/Heterotopic pregnancy were listed as the diagnostic hypothesis. Thus, MVA was performed with material sent to the pathological anatomical specimen, which presented a sample of decidualized stroma, blood and hydropic chorionic villi, consistent with complete hydatidiform mole and absence of malignant signs. On the fifth day of post-surgical hospitalization, the patient was discharged from the hospital and referred to the Regional Hospital of Asa Norte - DF, where there is a specialized service in the follow-up of patients with trophoblastic diseases. Comments: We observed the importance of serial laboratory follow-up of beta-hCG for the evolution of gestational trophoblastic disease, since when well managed, these women have a good prognosis. This patient also had a recent previous diagnosis of ectopic pregnancy, in which fertilization occurs outside the uterus. In the last

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three decades, there has been a greater number of cases of ectopic pregnancy, with an approximate fivefold increase. This is basically due to the higher incidence of PID, the more liberal use of the IUD, microsurgeries on the tubes and in vitro fertilization. Thus, heterotopic pregnancy, a rare condition, is a differential diagnosis that should be part of the specialist's arsenal, since the presence of a topical pregnancy does not exclude a concomitant ectopic pregnancy.

**Keywords:** Hydatidiform mole. Gestational trophoblastic disease. Ectopic pregnancy. Heterotopic pregnancy.



#### INTRODUCTION

Trophoblastic disease is a spectrum of rare gestational disorders related to the placenta and includes premalignant diseases, partial and complete hydatidiform mole, in addition to malignant ones, such as gestational trophoblastic neoplasia. Among these, hydatidiform mole (or molar pregnancy) is the most common disorder. They differ according to different cytogenetic, pathological, and clinical features, but in both a defect in gametogenesis or fertilization results in the presence of edematous placental villi. In addition, the conceptus is abnormal, carries two copies of the paternal genome and has an absent maternal genome (complete hydatidiform mole) or present (partial hydatidiform mole). In most cases, this disorder resolves after uterine evacuation, but it can persist and evolve into gestational trophoblastic neoplasia. (JOYCE et al., 2022; SOPER, 2021) Both types of molar pregnancies can follow this evolution, characterized by locally invasive or metastatic malignancy resulting from abnormal products of conception. (ALBRIGHT, et al., 2020) So, gestational trophoblastic neoplasia is a gestational disorder that originates histologically from trophoblastic cells of the placenta and can retire as an invasive mole, choriocarcinoma, trophoblastic tumor of placental location and epithelioid trophoblastic tumor. (CHEN, et al., 2022)

Ectopic pregnancy, in turn, is a common gestational disease that refers to the implantation of fertilized eggs outside the physiological gestational site, the uterus. Predominantly, this implantation occurs in the uterine tube, responsible for transporting eggs from the ovaries to the uterus, so in this case it can be called tubal pregnancy. In addition, a tubal molar pregnancy can happen, but there are few cases recorded in the scientific literature (D'ASTA et al., 2022; WENJING, HAIBO, 2022). In addition, it is possible to have a synchronous coexistence of an intrauterine pregnancy and an ectopic pregnancy, a pathological condition called heterotopic pregnancy. But this rarely happens in natural conception, it is usually related to the assisted reproduction technique (OANCEA, et al., 2020). In this context, heterotopic pregnancy is a special type of ectopic pregnancy, characterized by the implantation of embryos in 2 different sites, mainly intrauterine pregnancy combined with tubal pregnancy (SHENG, 2022).

Women with heterotopic pregnancies have a better prognosis regarding the continuity of intrauterine pregnancy and its preservation of reproductive function when evaluated early. In general, the prognosis of spontaneous heterotopic pregnancy is similar to that of an ectopic pregnancy. While that of the fetus, remains uncertain even after



treatment, with approximately 35% of miscarriages occurring during pregnancy. It is worth noting that there is an influence on the prognosis regarding the treatment used and the biological difference of each patient. (OANCEA, et.al; 2020). Thus, the guidelines do not take a position on the best management for intrauterine pregnancy combined with extrauterine pregnancy. The treatment takes into account the patient's desire to maintain the pregnancy, the experience of the attending physician and the resources available in each hospital unit; This therapeutic management is divided into surgical and conservative treatment. Surgical therapy includes the use of forceps, curettage, manual intrauterine aspiration, hysteroscopy, uterine artery embolization, laparoscopy, or laparotomy, depending on the extrauterine implantation site. Conservative management consists of the use of methotrexate (MTX), potassium chloride, sodium chloride in high concentration or glucose injections. Since, if there is preservation of the topical pregnancy or if it is a complicated patient, the effects of MTX and embolization of the uterine arteries are contraindicated. (SHENG, et.al; 2022)element. The literature recommends minimal intraoperative manipulation of the uterus in order to avoid injury to intrauterine pregnancy. In addition, the presence of hemoperitoneum directs immediate surgical therapy. (OANCEA, et.al; 2020). Conservative management, while conservative management is reserved for cases in which women do not have free fluid in the cul-de-sac of Douglas or peritonitis, without other clinical complaints, hemodynamically stable and without a documented embryonic pulse. The success rate for conservative therapy was 65.52%. Therefore, the most important thing in this type of management is to be performed in hospital conditions that are subject to constant hemodynamic and ultrasonographic monitoring and a surgeon on standby for cases of emerging indications. (MŁODAWSKI, et.al; 2023).

This study aims to emphasize the importance of diagnosing heterotopic pregnancy in order to decide on the best management that includes therapeutic success, fertility preservation and maintenance of intrauterine pregnancy.

#### CASE REPORT

A 12-year-old patient, with no history of morbidity, primiparous, smoker, blood type B +, denies previous surgeries, comes to the hospital accompanied by her mother, complaining of mild pain in the lower abdomen, especially in the right iliac fossa, and denied transvaginal bleeding. In addition, the patient had transvaginal ultrasonography (US) showing a 12-mm image in the left annex with fluid and a 7-mm yolk bladder.



On physical examination, the patient was in good general condition, flushed, eupneic, abdomen without palpable masses, absence of peritoneal irritation, and presence of pain on deep palpation of the hypogastrium. In addition, the vaginal examination showed a closed cervix that was painless on mobilization, without transvaginal bleeding and bulging. Thus, the hypothesis of tubal abortion was raised.

Thus, quantitative beta-HCG and a new ultrasound were requested, with results > 15,000 mIU/mL and an adnexal mass of 12 mm with no intrauterine alteration, respectively. In the aftermath, after hospitalization, the patient presented with pain on superficial palpation in the hypogastrium of strong intensity. As a result, an exploratory laparotomy was performed, at which time they identified the presence of a moderate amount of blood in the abdominal cavity, but normal fallopian tubes and ovaries bilaterally, absence of a gestational sac in the abdominal inventory, no inflammatory signs in the cecal appendix and the presence of thin pelvic adhesions. Thus, abdominal wall raffia was performed in planes, without surgical complications.

On the following day, the first postoperative day, the patient developed diffuse abdominal pain of moderate intensity without peritonism, did not evacuate, denied nausea or bleeding, and was hemodynamically stable. Moreover, the post-surgical examinations showed Hb 11.5 / Ht 34.3 / leukocytes 12,700 without left shift / Platelets 272,000, a new quantitative Beta-HCG is awaited. At this time, the patient was already monitored by the social service, guardianship council and police station of the city of origin.

Therefore, on the second postoperative day, the patient evolved with significant improvement in abdominal pain and bowel movements. With the result of the new Beta-HCG (>15,000 mIU/mL), presenting the same value as before, a new transvaginal ultrasound and total abdomen are requested with the following results: Acoustically normal liver, pancreas and spleen. A yolk bladder of normal shape, size, walls and contents, does not show stones inside. Right and left kidney were unchanged (total abdomen US). On the other hand, transvaginal ultrasound showed a flexed retroverse uterus measuring 7.6 x 5.8 x 6.2 cm, uterine cavity with echogenic endometrium, thickened of 27 mm, irregular and with anechoic areas intermingled, normal ovaries, and free posterior cul-de-sac. Therefore, gestational trophoblastic disease/Ovular remains/Heterotopic pregnancy were listed as the diagnostic hypothesis.

Thus, on the fourth postoperative day, the laboratory results showed Hb 12.6/ Ht 38/ 7000 leukocytes/ 311,000 platelets/ Creatinine 0.72/ AST 20/ TGP 13/ Urea 23, and she



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was referred to the operating room for MVA for uterine evacuation. Thus, during the procedure, several irregular segments of tissue were aspirated, burgundy, elastic-friable, measuring 8 x 8 x 4 cm, with multiple vesicles intermingled. The patient was sent to the pathological anatomical examination which presented a sample of decidualized stroma, blood and hydropic chorionic villi, consistent with complete hydatidiform mole and absence of malignant signs in the sections examined.

On the fifth day of post-surgical hospitalization, the patient was discharged from the hospital and referred to a Regional Hospital, where there is a specialized service for the follow-up of patients with trophoblastic diseases, for the result of chest X-ray and quantitative beta-HCG. In addition, monthly injectable contraceptives were prescribed in order to avoid a new pregnancy that would aggravate the current condition.

In view of the above, the minor continues in multidisciplinary care with social assistance, a specialized medical team, a guardianship council and psychology. This, in turn, detected a history of sexual violence practiced by the stepfather at the age of 6, the mother's current partner, in addition to emotional abandonment on the part of the mother, rejection of the relationship with a 17-year-old adolescent and a past of self-mutilation in the face of the family and social vulnerability in which she is inserted.

## **DISCUSSION**

In view of this case, we observed the importance of serial laboratory follow-up of beta-hCG for the evolution of gestational trophoblastic disease, since when well managed, these women have a good prognosis. Hydatidiform mole is a pathology where there is a fertilization error in the fertilization process of an egg without a nucleus and there is no embryo formation, in which it is divided into complete hydatidiform mole (MHC), in which the genetic material is completely of paternal origin (46 chromosomes). While, in Partial Hydatidiform Mole (MHP), it has a triple genetic load, due to the error of fertilization by different mechanisms, such as: dispermia, fertilization of a haploid egg by a diploid sperm or fertilization of a diploid egg by a haploid sperm.

Thus, the clinical evolution is based on the rupture of one or more vesicles associated with hyperemesis, ovarian cystosis, genital hemorrhage, and uterus enlarged for gestational age. Thus, the frequent use of ultrasonography and the quantitative measurement of b-hCG in pregnancy detect MH before the patient's bleeding. Trophoblastic



disease has early and advanced age as risk factors, and is common in adolescents and women over 40 years of age.

Concomitantly, this patient also had a recent previous diagnosis of ectopic pregnancy, in which fertilization occurs outside the uterus. In the last three decades, there has been a greater number of cases of ectopic pregnancy, with an approximate fivefold increase. This is basically due to the higher incidence of PID, the more liberal use of the IUD, microsurgeries on the tubes and in vitro fertilization. The diagnosis of ectopic pregnancy is not very predictable only by the clinic and even with parameters of complementary tests, such as b-hCG and ultrasound, the vast majority of cases are still diagnosed in the emergency, when the acute abdomen is installed by hemoperitoneum.

In addition, there is also heterotopic pregnancy, a rare condition in which there is an ectopic pregnancy simultaneous to an intrauterine pregnancy. It has an incidence ranging from 1:30,000, however after assisted reproduction the frequency of this complication went to 1:100-500 pregnancies. Good management of this condition interferes with women's prognostic and reproductive factors, as well as the continuity of intrauterine pregnancy, when feasible.

### CONCLUSION

Heterotopic pregnancy, a condition in which, even though it is rare, is a differential diagnosis that should be part of the specialist's arsenal, since the presence of a topical pregnancy does not exclude a concomitant ectopic pregnancy, even more so in cases of adolescents with clinical complaints of acute abdomen. Therefore, ultrasonography is the method of choice for the best evaluation of these patients. In addition to the evaluation of serial b-hCG dosage for strict follow-up and diagnosis of gestational trophoblastic diseases. Thus, aiming to preserve the fertility and survival of these women.



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