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## Case Report

# Ectopic molar pregnancy, rare entity: new case report

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## ABSTRACT

We report in this work the case of a 24-year-old patient who was admitted to the emergency room with an acute abdomen, the investigations were in favor of a ruptured EP, the patient underwent a salpingectomy the same day due to the state of the tube, the anatomopathological study of the product was in favor of an ectopic GTD, the patient benefited from a monitoring of the levels of BCHG according to the recommended protocols, we discussed through this case the different clinical and therapeutic aspects of this rare entity.

**Keywords:** Gestational trophoblastic disease, Ectopic pregnancy, Ectopic molar pregnancy, Monitoring, Anatomopathological study

## INTRODUCTION

Gestational trophoblastic disease (GTD) is a heterogeneous group of pregnancy disorders characterized by abnormal proliferation of trophoblastic tissue. It includes premalignant partial and complete hydatidiform mole, but also malignant invasive mole, choriocarcinoma, trophoblastic tumor of the placental implantation site and epithelioid trophoblastic tumor.<sup>1</sup>

We report a case of ectopic molar pregnancy a very rare entity in gynecology diagnosed at the stage of anatomopathological study on the salpingectomy piece, we will discuss through this case the different clinical and therapeutic aspects of this rare entity.

## CASE REPORT

Mrs B. S., 24 years old, gravida 2 para 1, with an antecedent of cesarean 4 years ago, currently she's seven weeks and six days pregnant according to a precise date of the last period, admitted to the emergency department for acute pelvic pain with medium abundance black vaginal bleeding.

### Physical examination

A conscious patient stable hemodynamically and respiratory, normo-colored conjunctivae was observed. On the gynecological level examination was (a) inspection: Pfan-type scarring; (b) abdominal palpation: localized defense in the left iliac fossa; (c) speculum: purplish cervix, bleeding from the endocervix; and (d) vaginal examination objectified anteverted uterus of normal size and, no laturo-uterine mass, positive Douglas' cry.

A pelvic ultrasound was performed showing a normal sized uterus, 16 mm thickened endometrium, a gestational sac with embryo and positive cardiac activity in the left latero-uterine area and thin effusion slide.

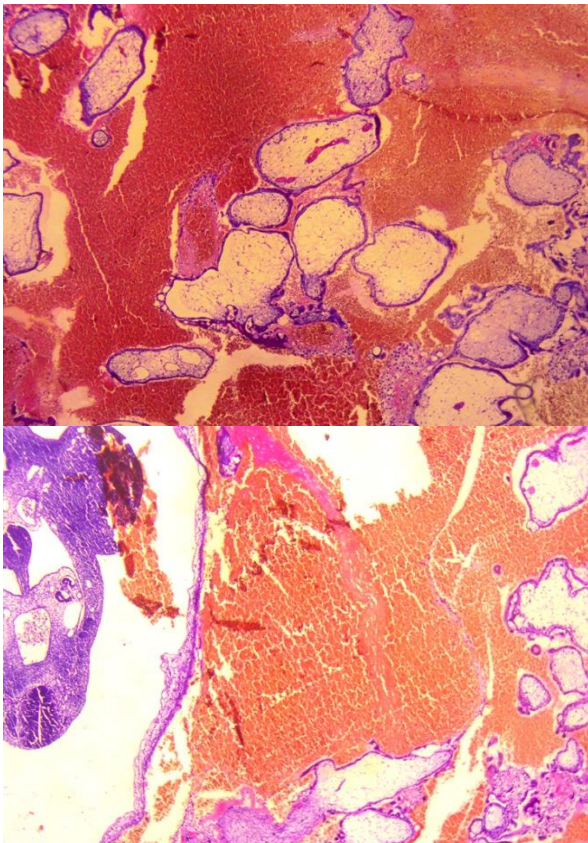
### Biologically

Biological study showed HCG was positive: 6520, hemoglobin and hemostasis test were corrects. Given the clinical, biological and ultrasound suspicion of an EP with signs of peritoneal irritation, the patient was referred to the operating room for exploration.

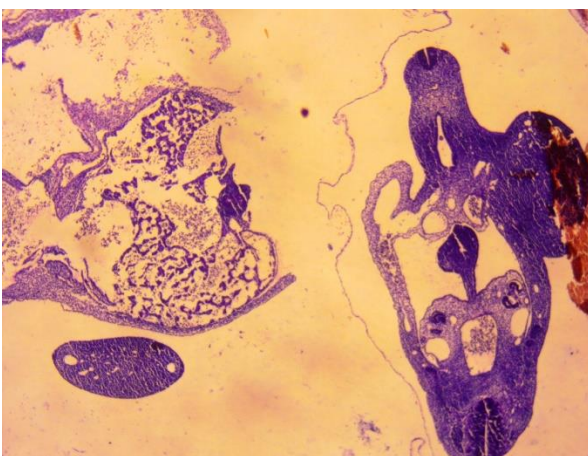
The patient underwent an exploratory laparotomy the same day, which showed a small amount of haemoperitoneum estimated at 200 CC aspirated with left EP, the patient underwent a retrograde left salpingectomy

The post-operative follow-up was simple.

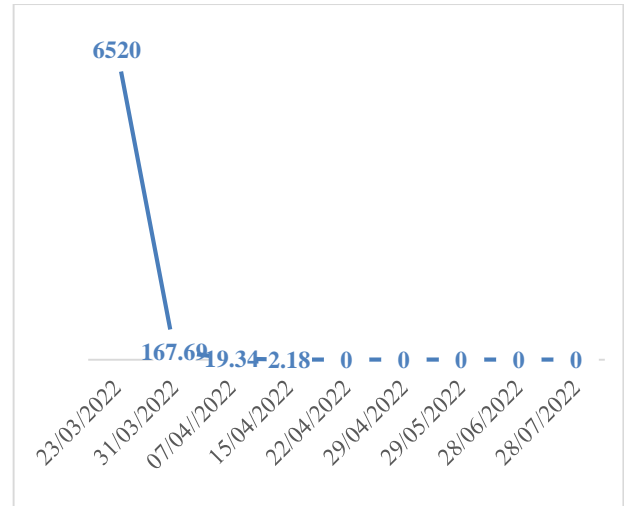
The histological examination of the uterine tube was in favor of an unruptured partial tubal mole with presence of embryo.



**Figure 1: Histological images showing avascular hydropic villi.**



**Figure 2: Histological image showing small fibrous villi with the embryo.**



**Figure 3: Curve showing the evolution of  $\beta$ HCG rates.**

## DISCUSSION

GTD is a heterogeneous group of pregnancy disorders characterized by abnormal proliferation of trophoblastic tissue. It includes premalignant partial and complete hydatidiform mole, but also malignant invasive mole, choriocarcinoma, trophoblastic tumor of the placental implantation site and epithelioid trophoblastic tumor.<sup>1</sup>

The anatomopathological study of the aspiration product is indispensable to confirm the diagnosis for a possible surveillance because there is a risk of malignant degeneration observed more in complete mole than partial mole.<sup>2</sup>

Among the risk factors of this pathology found in the literature are: extreme ages <20 years and >35 years (the risk is multiplied by 5 to 10 times in patients over 35 years), history of infertility or miscarriage, dietary factors (hypovitamin A, animal fat deficit), smoking.<sup>2,3</sup>

The metrorrhagia is the first motif of consultation but other symptoms can be seen as:

hyperemesis due to high levels of BHCG especially in case of complete hydatidiform mole, in more advanced cases extragynecological signs may be seen in relation to metastasis such as dyspnea, hemoptysis.

The discovery may be fortuitous.<sup>3</sup>

Pelvic ultrasound and BHCG is the gold standard couple to confirm the diagnosis. Intrauterine hydatidiform mole is usually diagnosed during a routine first trimester ultrasound.<sup>1,3</sup>

In case of complete hydatidiform mole it shows an enlarged uterus with a heterogeneous echogenic image with several hypoechogenic foci and multiple cystic spaces like grape cluster with no fetus.<sup>4</sup> sometimes ovarian

luteal cysts can be seen due to ovarian hyperstimulation by  $\beta$ HCG.

In partial mole, an elongated empty gestational sac or fetal parts (most often inviable) may be found. In rare cases of late gestational age pregnancies, growth retardation, oligohydramnios and an enlarged placenta with multiple diffuse anechoic lesions corresponding to cystic degeneration may be found.<sup>5</sup> HCG, is a specific tumor marker of the disease produced by hydatidiform moles and TTG it is much higher in case of complete hydatidiform mole compared to the gestational age range ( $>100000$ ).<sup>1,6</sup> It constitutes an excellent non-invasive means for surveillance, the diagnosis of GTT can be made even without anatomopathological evidence in the presence of abnormal variations in BHCG levels after evacuation (reascension, plateau), which allows a rapid and effective management.<sup>7</sup>

Pelvic CT and MRI are complementary examinations of locoregional and distant extension for advanced stage disease (GTT).<sup>1</sup>

In our case, the initial diagnosis was an Ectopic pregnancy with the following criteria: (a) clinical- metrorrhagia, signs of peritoneal irritation; (b) biological: Bhcg positive; (c) ultrasound: typical aspect of an EP (empty uterus + gestational sac with embryo and positive cardiac activity in the left latero-uterine area); and (d) context: cicatricial uterus.

The diagnosis of an ectopic mole was made at the stage of anatomopathological study on the salpingectomy piece, which is in agreement with the other studies: five cases of molar pregnancy of tubal location were reported in the study of rahaoui et al over a period of seven years. The patients were also referred to the operating room for suspected EP all patients received radical treatment. The diagnosis of hydatiform mole was obtained only after anapathological results the monitoring and evolution of the 5 patients were favorable.<sup>8</sup>

Over a period of 14 years 31 cases of ectopic mole were reported by Gillespie et al out of 5581 cases of gestational trophoblastic disease all patients presented with an acute abdomen.<sup>9</sup>

The anatomopathological diagnosis of ectopic molar pregnancy remains difficult because ectopic pregnancies may also present hydropic villi.<sup>10</sup> The study of DNA cytometry seems interesting to confirm the diagnosis.

In the absence of this confirmation the management will be the same as gestational trophoblastic disease (monitoring).<sup>11</sup>

The association of these two situations in gynecology is very rare, estimated at 1.5 cases/1,000,000 pregnancies and is described less than 300 times in reviews of the literature.<sup>9,12</sup>

We are in front of an exceptional case: given the reduced risk of the association of these 2 pathologies, absence of risk factor of molar disease

Our management was: retrograde salpingectomy for EP.

After the anatomopathological results, we completed by a weekly monitoring of  $\beta$ HCG until negatvation and then a monthly monitoring of the  $\beta$ HCG level for 6 months (Figure 1).

The management should be the same as intrauterine TTG, (evacuation of the product of conception salpingectomy/salpingotomy then monitoring of  $\beta$ HCG levels). Cases of ectopic TTG have been reported in the literature.<sup>5,13</sup>

The choice between conservative or radical treatment is very difficult to evaluate because the diagnosis of an ectopic mole is obtained only after surgery on anapath product.

In most studies the choice between the two surgical strategies depended on the local state of the uterine tube

## CONCLUSION

The association of GT disease and EP is a very rare entity, the preoperative diagnosis is difficult or even impossible and is only obtained after anatomopathological study, the picture is often an EP, the per-op suspicion is possible in case of ruptured EP with presence of vesicles, the management is the same as an intra-uterine gestational trophoblastic disease including an evacuation of the product and monitoring of BHCG according to the standard protocols to fight against any malignant transformation.

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