

# Intramural ectopic pregnancy

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## SUMMARY

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**Gestational trophoblastic neoplasia refers to a subset of gestational trophoblastic conditions characterized with persistently elevated serum  $\beta$ -human chorionic gonadotropin, absence of a normal pregnancy, and a history of normal or abnormal pregnancies. We described a case of suspected ectopic molar pregnancy in a primiparous woman who had elevated  $\beta$ -human chorionic gonadotropin and required chemotherapy to achieve remission. Final histopathological finding was ectopic pregnancy; no gestational trophoblastic neoplasia was found. This case stresses the importance of histopathological analysis in diagnosis of gestational trophoblastic neoplasia when ectopic pregnancy is present, considering that histopathological analysis is less sensitive for gestational trophoblastic neoplasia than for ectopic pregnancy.**

**KEY WORDS:** Pregnancy, Ectopic; Gestational Neoplasms; Diagnosis, Differential; Chorionic Gonadotropin, beta Subunit, Human; Antineoplastic Agents

## INTRODUCTION

Gestational trophoblastic disease (GTD) encompasses a broad spectrum of conditions that includes hydatidiform mole, invasive mole, placental site trophoblastic tumor (PSTT), and choriocarcinoma. Nowadays, GTD has become the most curable of gynecological malignancies for several reasons: (1) a sensitive marker is produced by the tumor,  $\beta$ -human chorionic gonadotropin ( $\beta$ -hCG) and the amount of hormone produced is directly related to the number of viable tumor cells; (2) this tumor is extremely sensitive to various chemotherapy agents; (3) risk factors for recurrence are known, allowing treatment to be individualized, and (4) the aggressive use of multiple treatment modalities, such as single- and multi-agent chemotherapy regimens, radiation and surgery (1,2). Appropriate chemotherapy and surgical treatment result in excellent survival (approaching 100%) and maintenance of fertility in the majority (80%) of women with post-molar gestational trophoblastic neoplasia (GTN) (3). However, a delayed diagnosis may increase the patient's risk of developing malignant GTN and adversely affect the response to treatment, and therefore the prompt identification of GTN is important (4). Raised serum  $\beta$ -human chorionic gonadotropin ( $\beta$ -hCG) apart from pregnancy or GTD, can occur because of menopause, a high normal level or a false-positive result. Accurate differentiation between these cases is vital to avoid potentially inappropriate investigations and therapies, which may induce infertility or other serious adverse events (4).

## CASE

A 32-years-old woman (one pregnancy, one birth delivery) was referred to Institute of Gynecology and Obstetrics, Clinical Center of Serbia in Belgrade because of elevated  $\beta$ -hCG (3500 mIU/ml), twelve days after induced abortion. Pelvic ultrasound (US) examination was performed which revealed suspicious residual tissue (Figure 1).

Curettage was repeated and the sample was sent to histopathological examination. The result was normal. During the follow-up period, in the second week, an increase in  $\beta$ -hCG level (26391 mIU/ml) was observed. Repeated pelvic ultrasound and magnetic resonance imaging (MRI) confirmed a presence of hyperechogenic mass in left uterine horn enclosed by serosa, and appeared to be in myometrium, suspected to trophoblastic neoplasia. The patient was admitted to chemotherapy. She was given intramuscular methotrexate 0.4 mg/kg daily for 5 days. She had

no complications during therapy. Over the next three weeks, level of  $\beta$ -hCG was checked on days 1, 7, 14 and 21. After twenty-one days, relevant decrease of  $\beta$ -hCG level was detected (263 mIU/ml), but the control US examination confirmed earlier findings and localization of neoplasia in left uterine horn (Figure 2).



Figure 1. Doppler effect above tumor in left uterine horn shows no distinct vascularisation



Figure 2. Hyperechogenic tumor in myometrium, reaching cavum uteri, few centimetres from perimetrium

Laparoscopic surgery was performed in order to confirm and treat GTN. The serum  $\beta$ -hCG level was 112 mIU/ml on the first postoperative day. Later on, histopathological analyses of operated material revealed ectopic pregnancy. This finding excluded molar pregnancy, but the patient was followed-up according to a scheme for molar pregnancy. The follow-up plan was for weekly urine samples until three normal levels of  $\beta$ -hCG were achieved and then controls monthly. First normal  $\beta$ -hCG was reached after two weeks. Control ultrasound was normal with no signs of any hyperechogenic mass in left uterine horn.

She was advised to refrain from getting pregnant until her  $\beta$ -hCG levels had been normal for six months. She was also advised to avoid using hormonal pills for contraception and to use barrier method instead.

## DISCUSSION

Intramural pregnancy is extremely rare and difficult to diagnose with less than 50 reported cases described in the literature (5). Because of the high rate of uterine rupture in most cases, hysterectomy is often necessary. In most of the cases in the literature, the diagnosis is established after total hysterectomy. The optimal medical management for this condition is unknown (6).

If intramural pregnancy is discovered before rupture, conservative (injection of potassium chloride into the gestation, systemic or local methotrexate injection), and surgical management should be considered (5). At first, we suspected invasive mole and our patient was given methotrexate. As the tumor and its localization remained the same after the therapy, the patient underwent surgery because intramural myoma was suspected. However, the results of histopathological examination revealed ectopic pregnancy. With this, we enabled the patient to have normal fertilization in future.

As our patient had two dilations and curettages, this case supports the role of previous uterine trauma as an etiologic factor for intramural pregnancy (7,8). Transvaginal ultrasonography and MRI were not able to diagnose this kind of ectopic pregnancy in this case. According to literature, only in one case, MRI established the diagnosis of ectopic pregnancy and the treatment was conservative with local injection of methotrexate (9).

To our knowledge, this was a rare case intramural pregnancy revealed after surgical treatment and which at first was suspected to gestational trophoblastic neoplasia (5,10). This case emphasizes the importance of applying strict histopathological criteria for GTD when a sample of ectopic pregnancy is analyzed, considering that this tool is less sensitive for GTD than for ectopic pregnancy.

## Conflict of interest

We declare no conflicts of interest.

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