CEOG Clinical and Experimental Obstetrics & Gynecology

A case report of a patient with high β-hCG levels after operation because of primary broad ligament pregnancy

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Summary

Introduction: A broad ligament pregnancy is an extremely rare condition and diagnosis is frequently missed and finally made during laparotomy. This is a case of a young patient with high serum beta-human chorionic gonadotropin (β-hCG) levels after operation because of broad ligament pregnancy. *Case Report:* A 31-year-old multipara complained of intermittent lower abdominal pain with vaginal bleeding for four months. A color ultrasonography revealed a cystic mass in the left attachment area, indicating an interstitial tubal pregnancy. However, trophoblastic disease could not be excluded. She accepted conservative treatment with methotrexate (MTX) at first, but observation showed that conservative treatment was slow and accompanied with liver function damage. Therefore, exploratory laparotomy was performed. Intraoperative situations and postoperative pathology confirmed broad ligament pregnancy. Her serum β-hCG was sustained at a high level for three months after operation. Her examinations of serum, CT, and ultrasonography could explain this situation. *Conclusion:* Primary broad ligament pregnancy refers to pregnancy where implantation of the fertilized ovum occurs directly between the two leaves of the broad ligament. The gravid substance was removed, however serum β-hCG could not gradually return to normal levels. This case should be followed-up closely to prevent adverse outcomes.

Key words: Beta-human chorionic gonadotropin (β-hCG); Broad ligament pregnancy.

Introduction

Broad ligament pregnancy is a rare condition and it refers to the growth of the gestational sac between the two leaves of the broad ligament. This type of pregnancy occurs when the gestational sac grows behind the peritoneum. Broad ligament pregnancy occurs in only 1/75 to 1/163 of ectopic pregnancy cases, or 1/183,900 of normal pregnancy cases [1]. Early diagnosis is critical and essential, because a catastrophic complication can occur because of placenta separation at any stage. The rate of maternal mortality has been reported to be as high as 20% [2]. An ultrasound is the most effective method for diagnosing an abdominal pregnancy. However, sometimes a MRI had to be resorted to confirm diagnosis even after clinical and sonographic evaluation by experienced specialists. Intraoperative situations and postoperative pathology is the gold standard to diagnose [3]. The authors report a multipara patient in whom the diagnosis was missed with high beta-human chorionic gonadotropin (β-hCG) levels after operation because of primary broad ligament pregnancy

Case Report

A 31-year-old patient was admitted to the hospital on November 4, 2013. The patient complained of intermittent lower abdominal pain with vaginal bleeding for four months. The patient's menstruation was usually irregular, with her last menstruation dating four months ago. When menstruation stopped for one month, she was diagnosed to be pregnant according to serum β-hCG levels. Color ultrasonography showed no gestational sac in her uterus. After three days, the patient experienced a small amount of vaginal bleeding. After re-examination, color ultrasonography showed a small gestational sac in her uterus. Progestin was administered to prevent miscarriage. After three days, the patient suffered from lower abdominal pain accompanied with nausea, vomiting, and more vaginal bleeding than normal menstrual flow. The patient then experienced intermittent lower abdominal pain with vaginal bleeding. On October 1, 2013, color ultrasonography performed at a local hospital showed a small amount of residual fluid in the uterus. Curettage was then conducted, without histopathological examination. However, intermittent lower abdominal pain with vaginal bleeding still occurred after the operation. On November 2, 2013, the patient went to a local hospital for β-hCG examination, and showed a result of 7,579 mIU/ml β-hCG. Color ultrasonography displayed a mass in the myometrium, indicating the possibility of trophoblastic diseases. On November 4, 2013, the patient went to Shandong Provincial Hospital to examine blood β-hCG levels, and showed a result of 9,576 mIU/ml β-hCG. Moreover, color ultrasonography revealed a cystic mass in the left attachment area, indicating an interstitial tubal pregnancy. However, trophoblastic disease could not be excluded (Figure 1). After the patient was admitted to the present hospital, routine blood, liver

Revised manuscript accepted for publication April 23, 2015

Clin. Exp. Obstet. Gynecol. - ISSN: 0390-6663
XLIV, n. 1, 2017
doi: 10.12891/ceog3061.2017

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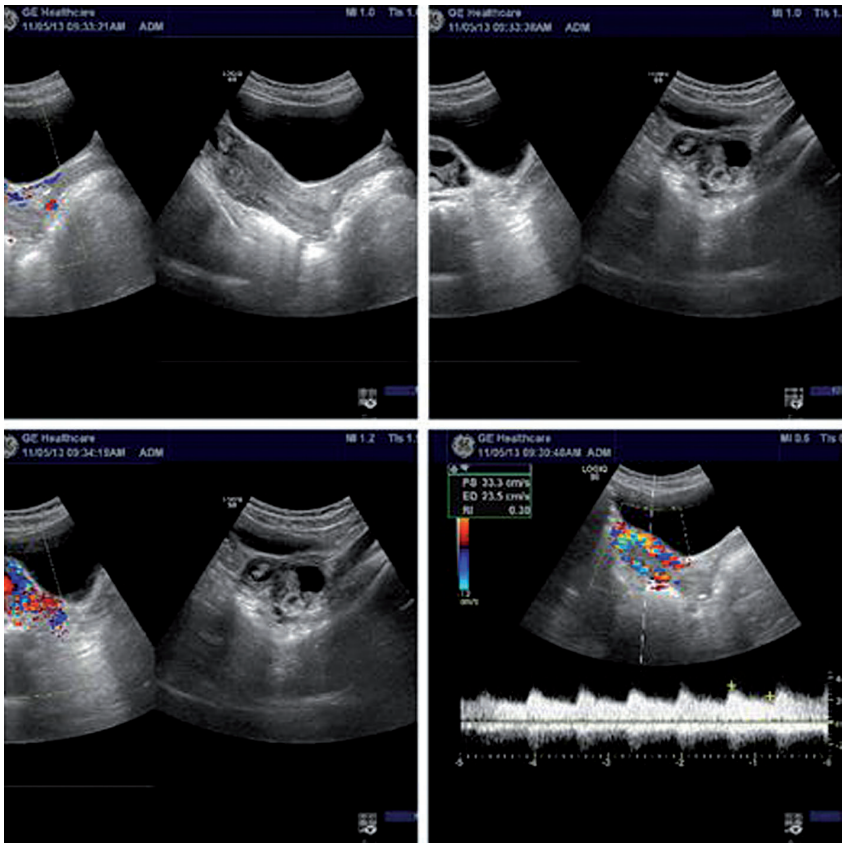

Figure 1. — Color ultrasonography performed on November 4, 2013.

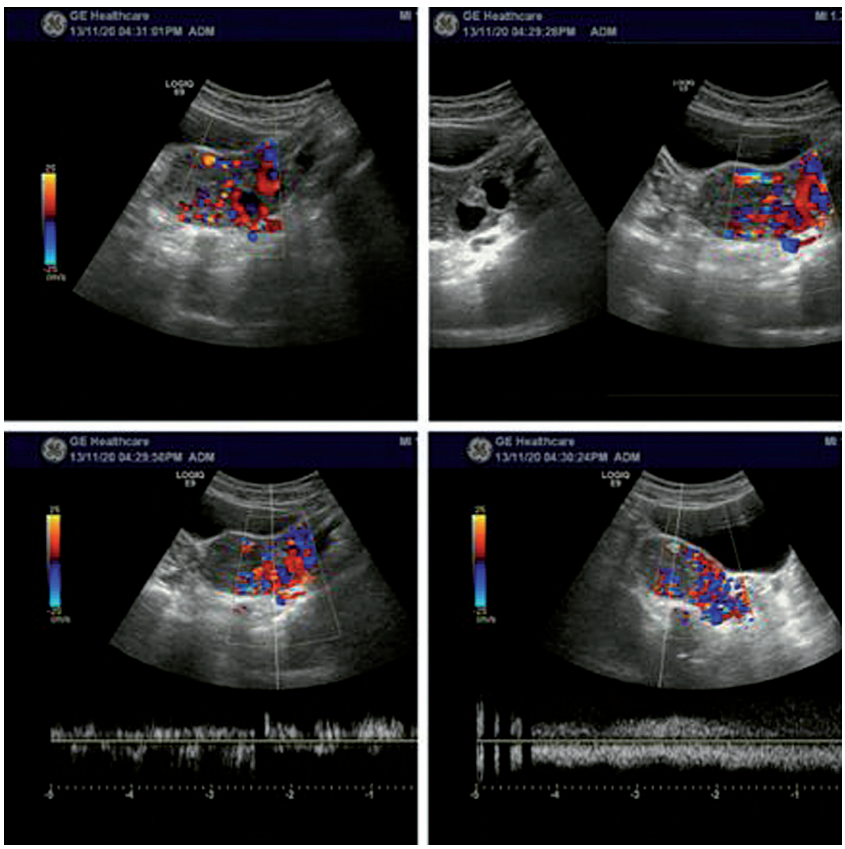

Figure 2. — Color ultrasonography performed on November 21, 2013.

function, biochemical tests, and CT of lung showed no obvious abnormality. The results of gynecological examination were as follows: (1) uterus was slightly large with an anteverted position and regular shape and felt tenderness; (2) a mass (diameter, of four cm) could be touched in the upper left side of the uterus in the left attachment area. The mass, which had a clear boundary and was closely related to the uterus, caused tenderness. Methotrexate (MTX) (50 mg qod) was administered three times by intramuscular injection. On November 2, 2013, β-hCG levels were re-examined, and a result of 1492.0 IU/L β-hCG was obtained and color ultrasonography had similar findings to the previous examination (Figure 2); MTX was continuously administered once again. On December 1, 2013, re-examination of β-hCG (680.7 IU/L) showed that liver function was abnormal, so liver-protective treatment was conducted. On December 6, 2013, re-examination of β-hCG (1038.0 IU/L) showed that conservative treatment was slow and accompanied with liver function damage. Therefore, exploratory laparotomy was performed on December 8, 2013. This procedure showed that a lump (diameter, of five cm) was found near the uterus in the left broad ligament, showing a violet-blue color in the surface and rich, twisted blood vessels in the surroundings. After the broad ligament anterior lobe serosa was opened, local thickening of blood vessel and a complete pregnancy sac (diameter of 1.5 cm) were observed. Bilateral fallopian tubes and ovaries were intact. When the vascular plexus was opened from inside of the left broad ligament, a large amount of blood leaked immediately. After peripheral vessels were sutured, MTX (ten mg) was injected from inside of the left broad ligament. Postoperative pathology showed villous edema and trophoblastic cells in the left broad ligament (Figure 3). After operation, re-examinations

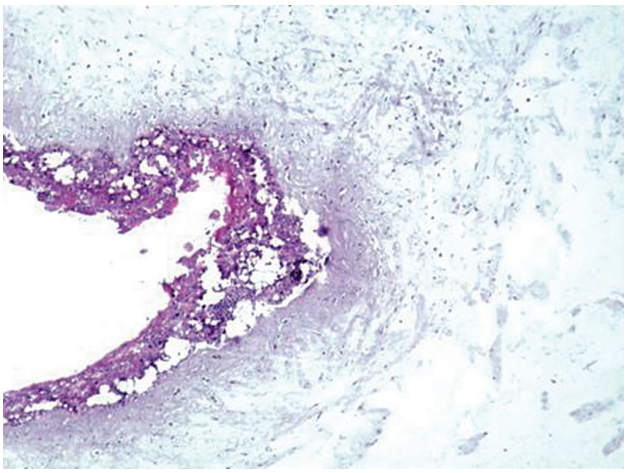

Figure 3. — Postoperative pathology.

on December 13, 2013 and December 16, 2013 showed β-hCG levels of 396.6 and 299.4 IU/L, respectively. Color ultrasonography, however, showed uneven myometrium echo and rich blood flow, which indicate gestational trophoblastic diseases (Figure 4). MTX (50 mg qod) was administered three times by intramuscular injection on December 17, 2013. On December 22, 2013, serum β-hCG was 106.6 IU/L. The patient was suggested to undergo blood re-examination until β-hCG levels were

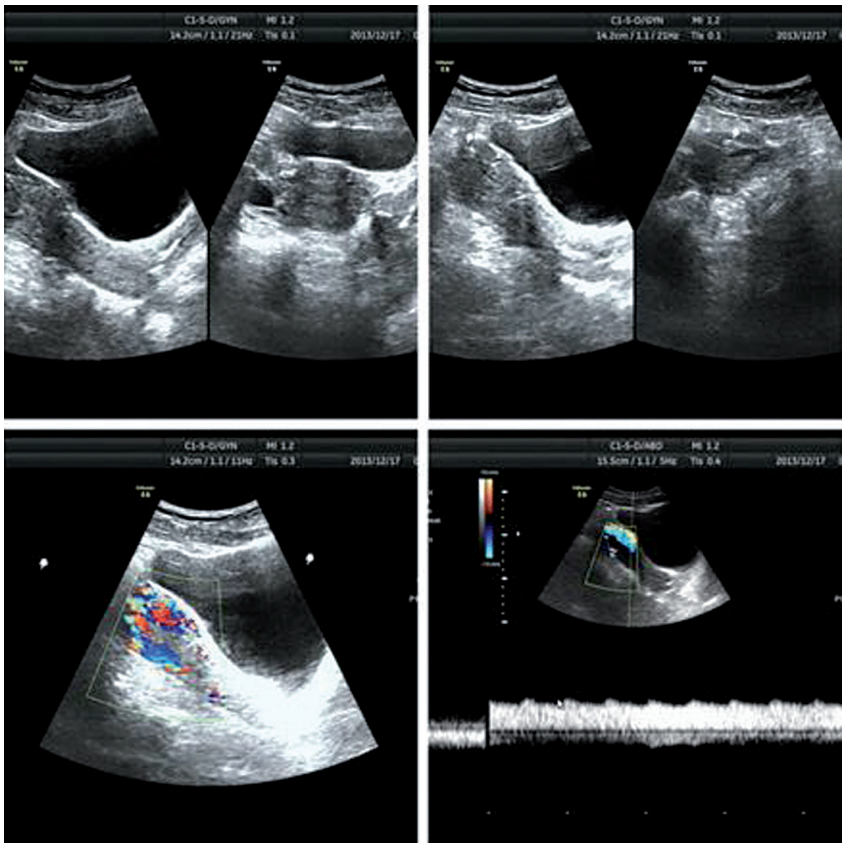

Figure 4. — Color ultrasonography performed on December 16, 2013 after surgery.

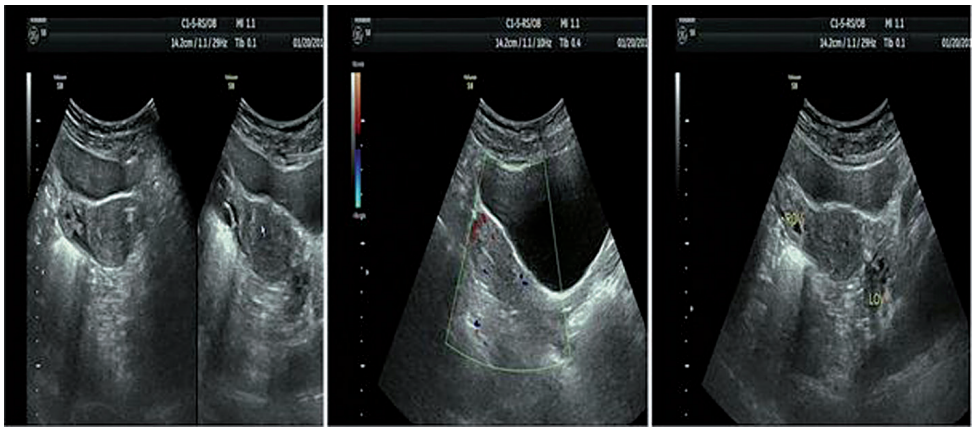

Figure 5. — Color ultrasonography performed on January 20, 2014.

within the normal range, and then was discharged from the hospital.

The patient had no sex after the operation, and underwent re-examination of β-hCG each week for three months. The re-examination results ranged from 76.0 IU/L to 191.50 IU/L, with slow growth each time. Color ultrasonography conducted on January 20, 2014 showed a middle strong echo on the bottom of the uterus measuring 1× .9×1 cm, without blood flow signal (Figure 5). After the operation, menstruation of the patient occurred normally, without abdominal pain and abnormal vaginal bleeding. On March 4, 2014, the patient's color ultrasonography showed a strong myometrium echo on the bottom of the uterus measuring 0.7×0.5×0.6 cm, without blood flow signal. Hence, a calcific focus may have been present.The patients was suggested to follow-up closely to observe her condition changes.

Discussion

A broad ligament pregnancy is divided into two main types. A primary broad ligament pregnancy refers to the fertilized ovum implant directly in the abdominal cavity. However, only 24 cases of primary abdominal pregnancy were reported up to 2007 [4]. In such cases,the fallopian tubes and ovaries are intact. A secondary broad ligament pregnancy accounts for most cases and it derives from tubal pregnancy penetrating trophoblasts through the tubal serosa and into the mesosalpinx, with transplantation between two leaves of broad ligament. It can also occur when developing a uterine fistula between endometrial cavity and the retroperitoneal space [5]. Under these circumstances, there is evidence of tubal or ovarian damage. In the present patient, the intraoperative situation confirmed a primary broad ligament pregnancy. A broad ligament pregnancy usually presents with mild abdominal pain or acute abdomen and shock after catastrophic internal haemorrhage. When patient does not present typical symptoms, clinical suspicion is often missed. The present patient complained of intermittent lower abdominal pain with vaginal bleeding for months with transient nausea and vomiting, which the authors thought was ordinary ectopic gestation.

Ultrasound is the most effective method for diagnosing ectopic gestation and MRI adds to diagnostic accuracy. MRI has been used to diagnose abdominal pregnancy [4]. However, as the diagnosis was not suspected, the present authors had not performed an MRI. The present patient was diagnosed with broad ligament pregnancy through surgical pathology. The gravid substance was removed so that blood β-hCG levels could gradually return to normal levels. However, weekly re-examination showed that the β-hCG levels of this patient slowly increased in a small range for three continuous months. Moreover, postoperative color ultrasonography indicated uneven tunica muscularis echo and rich blood flow. These results were possibly attributed to the following reasons: 1) partial trophoblast cells in the broad ligament pregnancy area moved to the tunica muscularis for survival proliferation, so color ultrasonography showed rich blood flow. After the gravid substance was removed in the operation, some trophoblast cells survived. Moreover, local injection of MTX did not kill all the trophoblast cells. After the operation, the remaining cells may continue to proliferate, causing high β-hCG levels. Hemorrhage may also occur after the operation, resulting in abdominal pain; 2) gestational diseases, a group of diseases from placental trophoblast cells, may develop. According to the histology, these diseases can be divided into vesicular mole, invasive mole, choriocarcinoma, placental site trophoblastic tumor, and epithelioid trophoblastic tumor. Histological classification of gestational trophoblastic diseases is necessary. However, invasive mole is similar to choriocarcinoma in clinical manifestation, diagnosis, and treatment principle. Gestational diseases often occur in young females who need to preserve their biological reproductive functions, with difficult acquisition of histological evidence. In 2000, the Society of Gynecologic Oncologist and, International Federation of Gynecology and Obstetrics suggested that the clinical classification of gestational trophoblastic diseases cannot be solely based on histology, and re-classification should be performed according to the range of lesion. Lesions confined to the uterus are called non-metastatic trophoblastic tumors, whereas lesions ap-

pearing at the site outside the uterus are called metastatic trophoblastic tumors. Postoperative color ultrasonography of the present patient showed uneven tunica muscularis echo and rich blood flow, so the possibility of trophoblastic diseases could not be excluded. All accessory examinations found no metastatic signs in the lungs and vagina. Therefore, the β-hCG levels of this patient should be continuously monitored. If the β-hCG levels continue to increase or if the rate suddenly increases, MTX treatment must be administered timely. Operation must be performed when necessary.

Several insights can be extracted from the evolution of the present case. The degree of cognition of this case was insufficient. Pathological examination should be made timely after diagnostic curettage in abnormal pregnancy. After MTX was injected intramuscularly many times, the preventive application of liver drugs was insufficient. Thus, liver function abnormality was found upon re-examination. MRI detection can be conducted for rare cases and those cases with poor results after multiple treatments. For the latter, doctors must consider other options. Furthermore, all accessory examinations must be performed carefully to avoid misdiagnosis and mistherapy.

Conclusion

This is a report of a patient with high β-hCG levels after operation because of primary broad ligament. The importance of this case report is the fact that a high β-hCG levels after conservative and operation treatment occurred for unknown reasons. In the present case, diagnosis of extrauterine pregnancy was made by imaging evaluation but final diagnosis of broad ligament pregnancy relied on laparotomy. Ultrasonography and clinical observations missed the diagnosis and MRI should be used as diagnostic tools to help the clinician in preoperative assessment when necessary in such challenging cases.

Acknowledgements

The authors are grateful to all of the study participants. They thank the help of the operating room, pathology department, image section, and intensive care personnel who assisted in the care of this patient. They also thank Edanz for assistance with English language editing.

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