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Case Report

Primary ovarian pregnancy: a report of four cases

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Abstract. Primary ovarian pregnancy is a rare form of ectopic pregnancy that constitutes 0.15-3 % of all ectopic pregnancies. Ovarian pregnancy results from the fertilization of trapped ovum within the follicle. Exact pre-operative diagnosis is often difficult, and with a few exceptions, the initial diagnosis is made on the operation table and the final diagnosis only through histopathology. The treatment of choice for ovarian pregnancy is usually ovarian wedge resection or oophorectomy, also there is a place for medical treatment of selected patients. Between October 2005 and May 2010, four cases of ovarian pregnancy that were detected and treated during laparotomy for suspected rupture of ectopic pregnancy are described. Three cases were treated by ovarian wedge resection. In one case, a single dose of 50 mg/m^2 methotrexate treatment was given, but the same mentioned surgical procedure were performed due to rupture of gestation. In all cases diagnoses were confirmed by pathologic examination. We aimed to discuss a rare form of ectopic pregnancy, primary ovarian pregnancy, and highlight the diagnostic criteria

Key words: Ovarian pregnancy, ectopic pregnancy

1. Introduction

The incidence of ectopic pregnancy was reported to be between 1-2% and fallopian tube was shown to be the most frequent location (1). Primary ovarian pregnancy is a rare form. It constitutes 0.15-3% of all ectopic pregnancies (2). The pathogenic mechanism consists of the fertilization taking place outside of the fallopian tubes and in the ovary. As a result of more widely used assisted reproduction techniques, in recent years, an increase in the rate of ovarian pregnancy was observed (3). In addition, the use of intrauterine instruments and sexually transmitted diseases are known to be factors responsible for ovarian pregnancy (4).

2. Case presentation

Case 1: A 21 years-old, G2 P0 A1 patient was admitted to the emergency room with severe abdominal pain. Abdominal examination of the patient revealed lower quadrant defense and sensitivity. During the vaginal examination, the uterus and both adnexa were sensitive to palpation. During the transvaginal ultrasonography, widespread free fluid in the pouch of douglas and in the adnexal area, and a 1.5 cm hyper-echoic mass image in the right adnexal area were observed. In the uterus, gestational sac was not observed. The result from the patient's serum β-hCG measured 4654 mIU/ml. At the pre-operative blood count of the patient, hemoglobin was 11 gr/dl and hematocrit was 33.6%. Based on these findings, the patient was taken to have an emergency operation due to the possibility of ruptured ectopic pregnancy. In the abdomen, approximately 200 cc of hemorrhagic fluid and coagulum were present. Both tubes and the left ovary appeared normal. On the right ovary, in a 2 cm area, actively bleeding ruptured ectopic pregnancy focus was observed. An approximately 2 cm wedge resection and full curettage was performed on the right ovary. The results from the pathology of the patient came back consistent with primary ovarian pregnancy. On the 3rd postoperative day, the patient was discharged without any

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complications and called back for periodic controls for serum β-hCG measurements.

Case 2: A 26 years-old G7 P5 A1 patient was admitted to the emergency room with severe abdominal pain. During the abdominal examination of the patient, a widespread sensitivity was present, especially in the lower quadrants. She had been using an intrauterine device for two years. Based on the preoperative blood count of the patient, hemoglobin was 12.5 gr/dl, hematocrit was 38.3% and serum β-hCG result was 564 mIU/ml. A gestational sac was not observed during transvaginal ultrasonography and right ovary was normal but left paraovarian liquid and free liquid in pouch of douglas was determined. The patient was taken in for operation with an acute abdomen diagnosis. In the abdomen, approximately 250 cc of hemorrhagic fluid and coagulum were present. Both of the fallopian tubes and right ovary were normal. On the left ovary, an actively bleeding ruptured mass with a size of 1cm was present (fig. 1). A 1.5 cm wedge resection and full curettage was performed on the left ovary. The results from the pathology of the patient came back consistent with primary ovarian pregnancy (fig. 2). On the 3rd postoperative day, the patient was discharged without any complications and called back for periodic controls for serum β-hCG measurements.

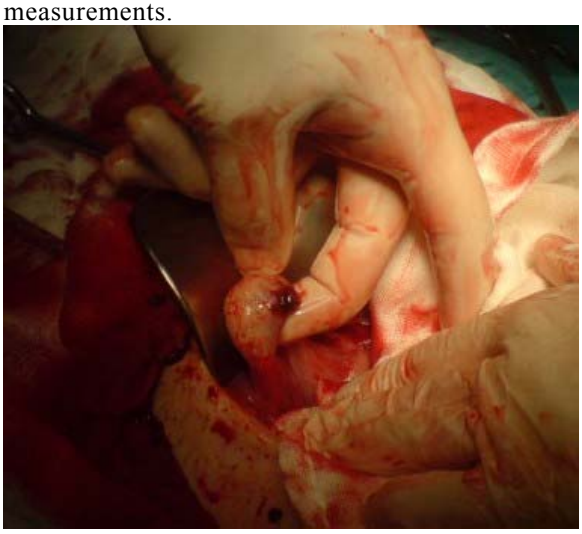

Fig.1. On the left ovary surface, a 1 cm-wide bleeding area due to ovarian pregnancy rupture is seen.

Case 3: A 38 years-old, G6 P2 A0 C3 patient was admitted to the emergency room with severe abdominal pain. During the abdominal examination of the patient, defense and sensitivity were present in the bilateral abdominal lower quadrant. She had been using an intrauterine device for four years. During the

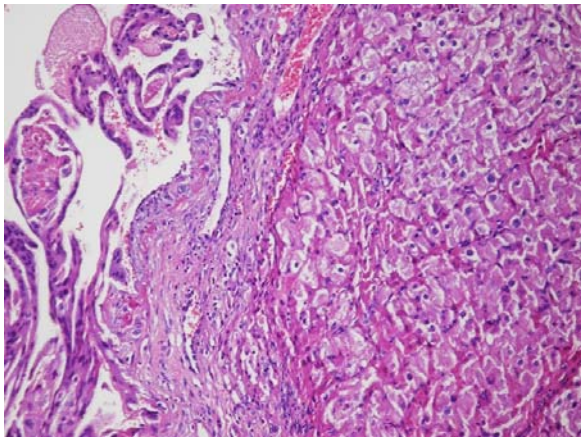

Fig. 2. Trophoblasts, contiguous to corpus luteum observed in ovarian tissue (HδEX200).

vaginal examination, the uterus and both adnexa were sensitive to palpation. During the transvaginal ultrasonography, widespread free fluid in the pouch of douglas and in both adnexal areas, and a 2 cm hyper-echoic mass image in the right adnexal area was observed. In the uterine cavity, gestational sac was not observed. The result from the patient's serum β-hCG measured 5071 mIU/ml. The preoperative blood count of the patient indicated that hemoglobin was 10 gr/dl and hematocrit was 30.6%. Based on these findings, the patient who was thought to have ruptured ectopic pregnancy was taken to have an emergency operation. In the abdomen, approximately 200 cc. of hemorrhagic fluid and coagulum were present. Both tubes and the left ovary appeared normal. On the right ovary, in a 2 cm area, actively bleeding ruptured ectopic pregnancy focus was observed. An approximately 2 cm wedge resection was performed on the right ovary. The results from the pathology of the patient came back consistent with primary ovarian pregnancy. On the 4th postoperative day, the patient was discharged without any complications and called back for periodic controls for serum β-hCG measurements.

Case 4: A 32 years-old G3 P1 A1 patient applied to our clinic with delayed menstrual period. The serum β-hCG result was 4117 mIU/ml. During the transvaginal ultrasonography, no gestational sac was observed in the uterine cavity and the endometrial thickness was measured to be 19 mm. Due to the presence of a 3 cm mass in the right adnexal area, she was hospitalized with the diagnosis of ectopic pregnancy. It was learned from the patient that she had two operations due to endometriosis and hydatid cyst. Medical treatment was performed using a single dose 50 mg/m^2 methotrexate

Table 1. Primary ovarian ectopic pregnancy cases

Case	Age	Obstetric History	Risk factor	Localization	β-hCG	Treatment
1	21	P0A1	Absent	Right	4654	Laparotomy-wedge resection
2	26	P5A1	IUD	Left	564	Laparotomy-wedge resection
3	38	P2A0C3	IUD	Right	5071	Laparotomy-wedge resection
4	32	P1A1	Pelvic surgery, endometriosis	Right	4117	Laparotomy-wedge resection due to rupture after methotrexate

protocol. On the 3rd day of the medical treatment, due to the development of abdominal pain and observance of free abdominal fluid in the ultrasonographic examination, emergency laparotomy was performed. In the abdomen, approximately 300 cc of hemorrhagic fluid and coagulum were present. Both tubes and the left ovary appeared normal. On the right ovary, in a 2 cm area, actively bleeding ruptured ectopic pregnancy focus was observed. An approximately 2 cm wedge resection of the right ovary and full curettage were performed. Pathological evaluation results were consistent with primary ovarian pregnancy. Our patient was discharged on the 5th postoperative day, without any complications. She was asked to return to our clinic for serum β-hCG measurements.

3. Discussion

The incidence of primary ovarian pregnancy shows similarities to interstitial pregnancy which is localized in the muscular layer of the uterus. Implantation to the ovary or the lack of follicular expulsion are the causes for ovarian ectopic pregnancies (3, 5). Ovarian pregnancies are classified as primary and secondary (4). Primary occurs if the ovum is fertilized while still within the follicle. Secondary occurs when fertilization takes place in the tube and the conceptus is later regurgitated to be implanted in the ovarian stroma (6).

While ovarian pregnancy may be related to risk factors such as; the use of intrauterine devices, reproductive technology, endometriosis, pelvic inflammatory disease and pelvic surgery, primary ovarian ectopic pregnancy may develop without any underlying risk factors (7,8). In two of our patients, there was a history of intrauterine device use, one patient had a history of endometriosis and pelvic surgery, however, in one case, there were no risk factors (Table 1).

Primary ovarian pregnancy has been defined by Spiegelberg using 4 criteria: the tube on the effected side where the pregnancy is must be intact, a gestational sac should be located in the ovary, gestational sac should be connected to the uterus by the utero-ovarian ligament and ovarian tissue must be surrounding the gestational sac. These are the required histopathologic criterias that cannot be verified by ultrasonography (1). In all of our 4 patients, the tubes were normal and an actively bleeding area was observed on the ovary. Based on these findings, all four of our cases meet the criteria. Whether it is during surgery or ultrasonography examination, it is hard to differentiate ovarian ectopic pregnancy from corpus luteum and hemorrhagic cyst. Ovarian pregnancies usually appeared on or within the ovary as a cyst with a wide echogenic outside ring. A yolk sac or embryo was less commonly seen (9). Attempts to use color or spectral Doppler ultrasonography to reliably distinguish a gestational sac from a corpus luteum have not been successful, because overlap with a corpus luteum has been high (10). Histologically, revealing trophoblast cells in the ovarian tissue confirms the ovarian ectopic pregnancy diagnosis (1).

Since ovarian pregnancy is generally diagnosed through laparoscopy or laparotomy, apart from possibly determining the diagnosis, laparoscopy is also the most common treatment approach: Either the entire ovary including the ectopic pregnancy has to be removed or a wedge resection of the ovary is necessary (1).

Due to ruptured ectopic pregnancy, our patients were treated by wedge resection through laparotomy. Since a rupture had not occurred during diagnosis, medical treatment of one patient was initiated through the administration of methotrexate. However due to rupture on the 3rd day of the treatment, laparotomy and wedge resection were conducted.

After taking into consideration the possibility that oophorectomy and wedge resection may decrease the ovarian reserve, Mittal et al. have treated ovarian pregnancy using laparoscopy assisted methotrexate injection (11). Injection of etoposide into the ovary is another option and it has fewer side effects (12).

If β-hCG values continue to be high after surgery, persistent trophoblastic disease should be considered and medical treatment should be conducted. High β-hCG level, high serum progesterone level, presence of cardiac activity (13), presence of yolk sac (14), high level of folic acid prior to treatment (15) and endometrial thickness >12 mm (16) are factors that complicate medical treatment. In one of our patient, the β-hCG value was 4117 mIU/ml and the endometrial thickness was 19 mm and was unresponsive to methotrexate treatment. In all of our cases, β-hCG values decreased gradually after surgery and persistent trophoblastic disease was not observed.

In conclusion, it is hard to reach primary ovarian ectopic pregnancy diagnosis prior to operation. In determining the treatment method, presence of rupture and vital signs of the patient are important factors. The priority of treatment should be to prevent mortality. If it is possible, fertility should be protected through conservative surgical methods or medical treatment of appropriate patients.

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