

A spontaneous bilateral tubal pregnancy

A case report

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Abstract

Rationale: Ectopic pregnancy (EP) is a condition in which a fertilized oocyte implants outside the uterine. Spontaneous bilateral tubal pregnancies are the rarest form of ectopic and are considered spontaneous when no fertility treatments are involved. There are few prior cases. Here I report a case of spontaneous bilateral tubal pregnancy.

Patient concerns: A 24-year-old female with long lasting vaginal bleeding of 29 days duration.

Diagnoses: Transvaginal ultrasound observed small complex masses on both adnexal sides without intrauterine gestational sac, and the hCG level was 21438 IU/L. But diagnosing based only on imaging findings and blood test result was difficult.

Interventions: Operative laparoscopy with salpingectomy bilaterally.

Outcomes: Villi were clearly shown in both resected tubes. A spontaneous bilateral tubal pregnancy was confirmed finally and the patient recovered uneventfully.

Lessons: Women of reproductive age with irregular menstruation should get access to diagnosis and appropriate treatment without delay. Salpingotomy should also be considered if the patient had a strong wish for future pregnancy. Developments of diagnosis and management of such condition are strongly called for.

Abbreviations: ART = assisted reproductive technology, EP = ectopic pregnancy, GS = gestational sac, hCG = human chorionic gonadotropin, IUP = intrauterine pregnancy, IVF-ET = in vitro fertilization embryo transfer, MTX = methotrexate, STD = sexually transmitted disease, TVUSS = transvaginal ultrasound scan.

Keywords: bilateral tubal pregnancy, case report, laparoscopy

1. Introduction

Although majority of ectopic pregnancy (EP) are implanted in the fallopian tubes, spontaneous bilateral EP remains a rare type of gestation. To date, only several published cases are available. The incidence of bilateral tubal pregnancy is only 1 out of every 200,000 spontaneous pregnancies^[1] and range from 1 out of every 725 to 1580 ectopic pregnancies.^[2] As salpingectomy bilaterally will lead to infertility naturally and no specific guideline has been developed to manage this condition, developments of diagnosis and management are strongly called for. The

case we presented here described a young girl who suffered from bilateral EP and underwent laparoscopy in the end. A further discussion of diagnosis and management on this matter is necessary.

2. Case report

A 24-year-old woman (gravida 3 para 1) who conceived naturally, attended the outpatient with vaginal bleeding of 29 days duration. A quantitative human chorionic gonadotropin (hCG) test was 21,438 IU/L, with an estimated gestational age of 7 weeks by dates. Transvaginal ultrasound was performed, which revealed complex adnexal masses measured 25 × 13 mm on the left side and a similar one measured 23 × 17 mm on the other side. In addition, no intrauterine gestational sac (GS) was visualized. Bilateral EP was suspected and she was admitted to the hospital. The abdomen was tender without rebound or guarding. Bimanual examination suggested uterine tenderness and bilateral adnexal tenderness. There were no obviously palpable masses. She denied syncope, dyspnea, nausea, or vomiting and did not have a history of pelvic inflammatory diseases or sexually transmitted diseases (STDs). She was hemodynamically stable with blood pressure of 124/85 mmHg, a pulse of 80 beats/min, temperature of 37.2°C, and oxygen saturation of 99% on room air. Because of the high level of hCG and for the benefits of less postoperative pain and fewer complications, a diagnostic laparoscopy was immediately done to inspect her uterus and adnexa, and masses were observed on both sides (Fig. 1). The patient indicated no desire to future pregnancy before operation; we decided to do salpingectomy bilaterally finally. Villi were revealed clearly in resected fallopian tubes (Fig. 2). The diagnosis

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Ethical review: The record or information of the patient was anonymized. Informed consent was obtained from the patient for publication of this case report.

The author has no conflicts of interest relevant to this article.

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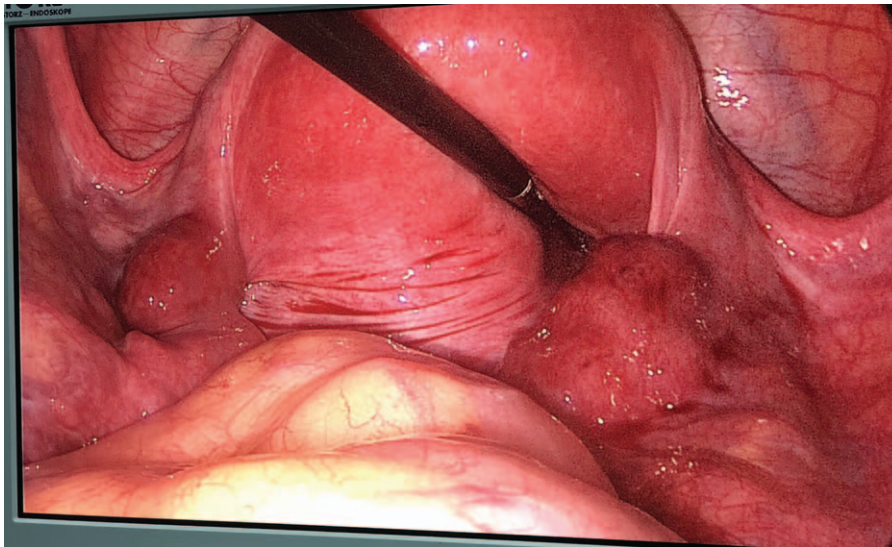


Figure 1. Adnexal masses on both sides.

of bilateral tubal pregnancy was confirmed. Serum hCG level declined sharply afterwards and the patient discharged without incidence 5 days later.

3. Discussion

EP is a common emergency in gynecological department. It occurs in approximately 1% to 2% of all pregnancies, and >98% implant in the fallopian tube.^[3] Risk factors include damage to the endosalpinx produced by STD, distortion of pelvic anatomy

caused by diseases such as endometriosis, or by adhesions after previous interventions, etc.^[1] Some investigations suggest that EP commonly occurs in women with multiple sexual partners, tobacco smoking, first intercourse at early age, impaired tubal function, vaginal douching, etc.^[4] Regarding the mechanism of bilateral EP, there are 4 proposed theories as follows: the occurrence of multiple ovulation spontaneously or by ovulation induction has increased the risk of bilateral EP^[5]; Ijland et al^[6] put forward the theory of the rhythmic endometrial wave-like movements in healthy normal cycling women. Correctly placed intrauterine fertilized ovum possibly migrates into fallopian tube as a result of the retrograde action of uterine section or contractions, even there are no operations of assisted reproductive technology (ART) and ovulation stimulation; women who use a progesterone-only pill or a progesterone-containing intrauterine contraception device, which will give rise to an elevated progesterone concentration, are more likely to suffer from EP, on account of the alteration of the circular muscular activity of fallopian tubes that is related to the abnormal progesterone concentration^[7]; transfer more than 1 embryo directly into fallopian tubes occurs during the operation of in vitro fertilization embryo transfer (IVF-ET), the inherent qualities of the transfer medium and a larger volume of transfer medium used probably have an impact on the occurrence rate of EP.^[8]

For women who suffer from EP, wide range of clinical presentations in early pregnancy results in difficulty in diagnosing in time. It severely threatens reproductive women's lives owing to unexpected intraperitoneal bleeding, hypovolemic shock, and lack of timely intervention in spite of technological advances allowing routine diagnosis of EP before clinical symptoms arise. Although in recent years, morbidity of bilateral EP has increased with the development of ART such as IVF-ET, spontaneous ones remain rare cases. Although transvaginal ultrasound scan (TVUSS) has a sensitivity of 87% to 99% and specificity of 94% to 99%, with positive predictive value of 96.7% and negative predictive value of 99.4% for identification of EP,^[3] in almost all the cases, the condition of bilateral EP has not been recognized until the time of surgery because even the most experienced sonographers may only see a handful during their lifetime. Thanks to the sufficient skills of our sonographer, we



Figure 2. Villi in both resected tubes.

were lucky to think of bilateral EP initially. Presentation of villi in resected specimens is a direct evidence; otherwise, pathological examination is a criterion standard as well if the surgeons fail to detect the villi visually.

Clinicians should be aware of the different management options and the patient's preferences are also of importance. The therapeutic effect of conservative treatment using methotrexate (MTX) is not satisfactory due to the high level of hCG (the hCG concentration indication for MTX therapy was <5000 IU/L).^[9] If the patient has a strong wish to preserve fertility, salpingotomy should be taken into consideration. Of course, patients should be informed of the possibility of incomplete removal of trophoblastic tissue and the risk of persistent trophoblast, additional treatment with systemic MTX, and repeat EP.^[10,11] Patients may focus on the fertility rates in relation to salpingotomy versus salpingectomy, according to Yao and Tulandi,^[12] subsequent fertility were similar with the 2 approaches. Likewise, in a randomized controlled trial, Femke et al^[11] suggested that cumulative rates of ongoing pregnancy by natural conception were similar between the 2 approaches. Because every study has limitations, women with a strong preference for future pregnancy might still opt for salpingotomy. In our case, consensus had been reached preoperatively and the surgeons did what they had agreed to the patient.

It is important for clinicians to understand the limitation of hCG.^[13] Although with the use of TVUSS, the "discriminatory zone" level of serum hCG is between 1000 and 1500 IU/L³, which describes the lowest hCG level in which ultrasound should detect visible signs of early pregnancy, such as fetal pole or yolk sac,^[14] not every patient will demonstrate similar findings on imaging test. Some investigators suggested several factors contributed to the nonvisualization of pregnancy, such as adenomyosis and uterine fibroids. They conclude the combination of serum hCG level and TVUSS findings is not predictive of eventual intrauterine pregnancy (IUP) or EP.^[15] In our case, the high serum hCG level had put the patient at high risk for EP due to no visible IUP and small masses without GS on both adnexal sides. When serum hCG levels are >2000 IU/L, the use of curettage to obtain the final diagnosis is particularly important.^[16] hCG concentration decreases not $<15\%$ after curettage in 8 to 12 hours is diagnosis of complete abortion; if hCG concentration does not fall, EP is diagnosed.^[17] Evidence on imaging test in our case cannot fully convince us to determine the location of fertilized ovum. Among women who were subsequently diagnosed with EP, 71% had the appearance of small heterogeneous masses.^[18] Caution is needed when taking a laparoscopic view only on basis of high serum hCG level and small adnexal heterogeneous noncystic masses without intrauterine GS. A computed tomography or magnetic resonance imaging may be needed in case of circumstances in which the EP may lie outside the scan range of the TVUSS^[3] such as retroperitoneal EP reported by Yizhuo et al.^[19]

In conclusion, even if unilateral EP is considered before operation, a close inspection of the abdomen and pelvis must always be done when performing surgery for an ectopic gestation, especially the contralateral fallopian tube. In several cases, the contralateral pregnancy was found days to weeks after the initial surgery.^[20] It is equally important to check both adnexal sides by TVUSS. Improvements in diagnosis are in progress; more and more special biomarkers have been discovered in recent years, for

example, according to Kiyonori et al,^[21] the concentrations of cell-free pregnancy-associated microRNAs (miR-515-3p, miR-517a, miR-517c, miR-518b) in maternal plasma were confirmed to be statistically significantly different in women with EP. Combination of the patient's complains, TVUSS results, serum hCG levels, and cooperation of doctors and sonographers are definitely of great importance. Being alert to EP in all women of reproductive age will allow more rapid diagnostic approaches and improve individualized management in the future.

Author contributions

Writing – original draft: Hong Xu.

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