Bilateral Ovarian Pregnancy After in Vitro Fertilization and Embryo Transfer in a Patient with Tubal Factor Infertility

Myoungseok Han,^{1,3} Jihyun Kim,¹ Hyunho Kim,¹ Goohwa Je,¹ and Taiyoung Hwang²

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Primary ovarian pregnancy is very rare event after natural pregnancy or assisted reproductive technology (ART) procedures. Although there are a few reports about unilateral ovarian pregnancy after in vitro fertilization and embryo transfer (IVF-ET), there has been no report about bilateral ovarian pregnancy. Moreover, it is difficult to diagnose an ovarian pregnancy following in vitro fertilization and embryo transfer because of enlarged ovary, fluid collection in pelvic cavity, and its low incidence. We present a case of a patient who underwent IVF-ET due to tubal factor infertility, but the patient developed bilateral ovarian pregnancy and was performed both ovarian wedge resection through laparotomy.

KEY WORDS: Both tubal obstruction; embryo transfer; in vitro fertilization; ovarian pregnancy.

INTRODUCTION

Ectopic pregnancy is well known complication after assisted reproductive technology (ART) procedures. However, primary ovarian pregnancy is very rare form of ectopic pregnancy following natural conception or in vitro fertilization and embryo transfer (IVF-ET). The reported incidence of natural primary ovarian pregnancy is 3.3% of all ectopic pregnancies (1). The incidence of primary ovarian pregnancy after IVF-ET is reported to be 0.27% per clinical pregnancy (2).

Although there has been several reports about primary ovarian pregnancy after IVF-ET, bilateral ovarian pregnancy in a patient with both tubal obstruction has not been reported until now. We report a case of bilateral ovarian pregnancy after IVF-

ET treatment for secondary infertility due to tubal factor.

CASE REPORT

The patient was 38 years old. She had undergone an operation of tubal reanastomosis 3 years ago. Eight months after the operation, laparoscopic left salpingectomy was performed due to left tubal pregnancy. At that time, there were somewhat adhesions with omentum at the distal part of right tube. But, we did not confirm the patency of right tube because both tubal patency had been presented at the time of the tubal reanastomosis and the right salpinx was intact grossly.

The hysterosalpingography (HSG) was performed because she did not conceive for 2 years since previous left tubal pregnancy. The HSG did not show the right tubal patency and we decided to perform IVF-ET. Diagnostic laparoscopy was not performed because she did not agree to that.

Controlled ovarian hyperstimulation was achieved using pituitary suppression with short protocol. Eight

¹ Department of Obstetrics and Gynecology, Dong-A University School of Medicine, Dongdaeshindong 3 Ga-1, 602-715, Seo-gu, Busan, Korea.

² Department of Obstetrics and Gynecology, Areum Women's Hospital, Busan, Korea.

³ To whom correspondence should be addressed; e-mail: hmsobgy@daunet.donga.ac.kr.

oocytes were retrieved by transvaginal sonographic aspiration. On day 2 of culture, three embryos of the four-cell stage were transferred.

Serum β -hCG was measured by RIA serially. Three weeks after embryo transfer, the serum β -hCG level was increased to 5602 mIU/mL. But, transvaginal sonography exam revealed only slight dilated endometrial cavity without definite gestational sac. Then, we thought that it might be an ectopic pregnancy, but we decided to exam again 1 week later. Repeat transvaginal ultrasound exam was checked 1 week later, but we found only a questionable gestational sac without any abnormality in pelvic cavity. Again, 1 week later we did not find a definite gestational sac except for an irregular and thickened lining within endometrial cavity. The bilateral ovaries were only cystic state without any finding of ectopic pregnancy. At that time, the serum β -hCG level was as high as 62,520 mIU/mL. For the diagnostic purpose, dilatation and curettage was performed and the pathologic report was only Arias-Stella reaction without villi. Confirming ectopic pregnancy, immediate surgical intervention was performed under the impression of ectopic pregnancy. At laparotomy, about 50 mL of bloody fluid was collected in the pelvic cavity and the right salpinx was intact grossly. But, there were darkish hemorrhagic bilateral ovarian cysts which were overriding each corpus luteum cyst. We resected both cysts by preserving normal ovarian tissue, and the right salpinx was also removed. The pathologic diagnosis was considered and bilateral ovarian pregnancy was confirmed.

DISCUSSION

Primary ovarian pregnancy is an uncommon form of ectopic pregnancy after IVF-ET or natural conception. Marcus and Brinsden (2) reported that the incidence was 6%: 8 primary ovarian pregnancies among 135 ectopic pregnancies after IVF-ET. But the incidence after natural conception is much lower, ranging from $\sim 1/60,000$ to $1/1,500$ deliveries, accounting for up to 3.3% of all ectopic pregnancies (3). Even though IVF-ET has been associated with a higher risk of primary ovarian pregnancy, there has been no report about bilateral ovarian pregnancy on the literature up to now.

Diagnosis of primary ovarian pregnancy is very difficult because of its rarity and asymptomatic state before rupture. Although the combination of quantitative serum β -hCG and transvaginal sonography increases the diagnostic accuracy of ectopic

pregnancy, the diagnosis of primary ovarian pregnancy mainly depends on physician's suspicion and experience. Actually, about 50% of primary ovarian pregnancies after IVF-ET might be diagnosed at an asymptomatic state (4). As a rule, if the serum β -hCG level is >1000 IU/L without a gestational sac in the uterus by the transvaginal sonography, the patient is regarded as having an ectopic pregnancy. At first, we considered this patient to have a tubal pregnancy or a heterotopic pregnancy or abortion. But on laparotomy, we could not find any pathologic lesion at the right salpinx or pelvic cavity except for some blood collection in Douglas pouch. And, both ovary showed irregular and enlarged hemorrhagic cysts overriding corpus luteums without active bleeding.

The mechanism of primary ovarian pregnancy after IVF-ET is indefinite, especially in a patient diagnosed with both tubal obstruction. In this case, it could be considered that the transferred embryo migrate to the left ovary through the right salpinx. Because this assumption also corresponds to the patient with tubal patency, this patient might not be applicable. Although right tubal obstruction has been diagnosed through HSG in this case, the possibility of tubal patency could be present because HSG might diagnose the obstruction erroneously. According to one study, two-thirds of patient with proximal tubal occlusion on HSG had laparoscopic evidence of normal salpinx with patency by methylene blue insufflation (5). Marcus and Brinsden (4) also reported three ovarian pregnancies that occurred on the side on which the tube was either blocked at the cornual end or absent, and the contralateral tube was the patent one. Therefore, bilaterality in this case can be explained by intra-abdominal migration of the embryo through the patent salpinx. The other possible mechanism of primary ovarian pregnancy is intrafollicular fertilization. But this is not applicable to this IVF-ET patient.

Preservation of ovarian tissue is the clinical goal of treatment, which has primarily been surgical. The preferred therapeutic procedure is ovarian cystectomy or wedge resection. We performed both ovarian wedge resection preserving normal ovarian tissue.

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