

# Ovarian pregnancy after bilateral salpingectomy in a patient with *in vitro* fertilization: a case report

Journal of International Medical Research

2022, Vol. 50(9) 1–7

© The Author(s) 2022

Article reuse guidelines:

[sagepub.com/journals-permissions](https://sagepub.com/journals-permissions)

DOI: 10.1177/03000605221123683

[journals.sagepub.com/home/imr](https://journals.sagepub.com/home/imr)

Mingle Zhang<sup>1</sup>, Caijun Zhao<sup>2</sup>, Jiahua Zheng<sup>1</sup>,  
Zhongkang Li<sup>1</sup>, Meiyun Yin<sup>1</sup>, Haiyan Li<sup>1</sup>,  
Yuan Wang<sup>1</sup> and Xianghua Huang<sup>1</sup> 

## Abstract

Ovarian pregnancy is rare but may occur with *in vitro* fertilization-embryo transfer in women who have undergone bilateral salpingectomy. We report a case of an approximately 30-year-old woman who had *in vitro* fertilization and a history of bilateral salpingectomy, and was diagnosed with an ovarian pregnancy. Laparoscopic enucleation of the gestational product in the ovary and ovarian remnant reconstruction were performed. The patient recovered well after surgery and was discharged home 5 days postoperatively.  $\beta$ -human chorionic gonadotropin was undetectable 3 weeks after the surgery. Awareness of the possibility of ovarian pregnancy after *in vitro* fertilization-embryo transfer is the most important step in an early diagnosis and treatment. Salpingectomy should be carefully performed to eliminate the risk of heterotopic pregnancy, especially in cases where a subsequent gestation is desired.

## Keywords

Ovarian pregnancy, *in vitro* fertilization-embryo transfer, bilateral salpingectomy, tubal pregnancy,  $\beta$ -human chorionic gonadotropin, fallopian tube

Date received: 16 January 2022; accepted: 15 August 2022

## Introduction

Ovarian pregnancy (OP) is one of the rarest forms of extrauterine pregnancy, accounting for 1/7000 to 1/40,000 pregnancies.<sup>1</sup> OP after *in vitro* fertilization-embryo transfer (IVF-ET) with a previous bilateral salpingectomy is even rarer, and only a few

<sup>1</sup>Department of Obstetrics and Gynecology, The Second Hospital of Hebei Medical University, Shijiazhuang, Hebei Province, China

<sup>2</sup>The Second Hospital of Hebei Medical University, Shijiazhuang, Hebei Province, China

### Corresponding author:

Xianghua Huang, Department of Obstetrics and Gynecology, The Second Hospital of Hebei Medical University, No. 215 West Heping Road, Shijiazhuang, Hebei 050000, China.

Email: [Huangxh2003@163.com](mailto:Huangxh2003@163.com)



Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative

Commons Attribution-NonCommercial 4.0 License (<https://creativecommons.org/licenses/by-nc/4.0/>) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (<https://us.sagepub.com/en-us/nam/open-access-at-sage>).

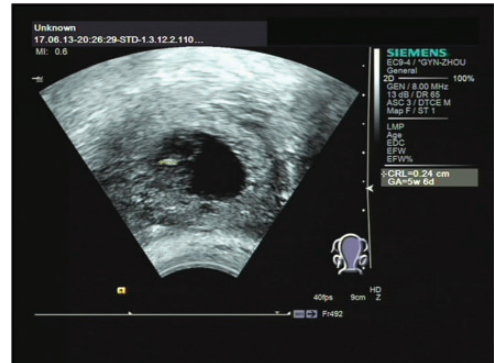
cases have been reported.<sup>2-5</sup> We report an ovarian pregnancy following bilateral salpingectomy for tubal pregnancy twice in a patient who had IVF. We also performed a literature search for related cases and discuss these cases.

## Case report

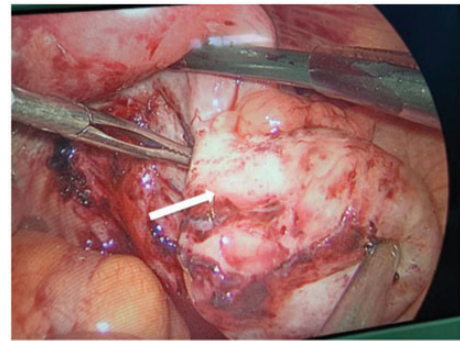
### Case presentation

A woman who was almost 30 years old, G3P0, with a history of a tubal pregnancy twice, presented to the Emergency Department complaining of minor lower abdominal pain, which started 1 day before her presentation. She was pregnant as a result of an IVF cycle. The patient had her first spontaneous pregnancy in her early 20s, and it was an ectopic pregnancy at the left fallopian tube and was treated with salpingectomy. Four years later, she became pregnant and the pregnancy was an ectopic pregnancy in the right fallopian tube, which was treated with salpingectomy. Three years later, the decision was made to treat her infertility via IVF after the surgery. This treatment was carried out and an IVF cycle was started. Twenty-three mature oocytes were collected and 16 were fertilized. Twenty-nine days before her admission to the Emergency Department, two thawed embryos on day 3 were transferred. Intramuscular progesterone in oil 60 mg daily was prescribed as luteal support. Her serum  $\beta$ -human chorionic gonadotropin concentration was measured 2 weeks after the transfer, and it showed a positive result. However, a transvaginal ultrasound showed an empty uterine cavity and a right adnexal mass, which strongly suggested an ectopic pregnancy (Figure 1). Her serum  $\beta$ -human chorionic gonadotropin concentration was 55,523 IU/L.

An injection of methotrexate was initially planned, but not performed to avoid bleeding due to puncture. An emergency



**Figure 1.** Ultrasound scan shows a gestational sac in the right ovary.



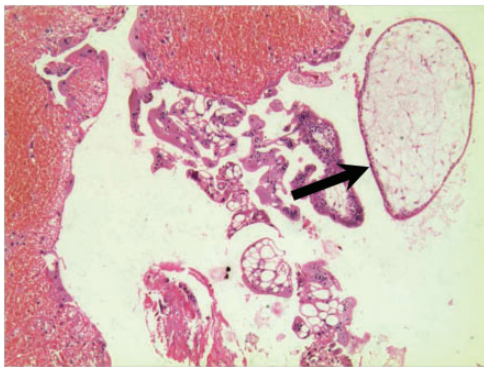
**Figure 2.** Intraoperative image of the pelvic area during laparoscopy. A slightly enlarged right ovary with a normal appearance can be seen with no right fallopian tube. The arrow indicates the location of the gestational sac in the ovary.

laparoscopy was performed after the patient provided consent for treatment. The operation showed thickened inflammatory walls of the stump of the fallopian tubes with adhesions to adjacent tissues, a slightly enlarged uterus, absent fallopian tubes, and a normal sized left ovary. The right ovary was slightly enlarged with a normal appearance (Figure 2). We found that the stump of the right tube was covered with the ovary because the adhesion between the uterus and the right ovary was separated. When a knot on the stump

of the right fallopian tube was pulled out, a ruptured gestational sac was observed in the ovary. Because this morphological feature suggested an ectopic right OP, wedge resection of the ovary at the site of abnormality was performed. The ovarian remnants were reconstructed with a 3/0 Vicryl suture. Approximately 600 mL of blood was lost during the operation. A pathological evaluation showed ovarian tissue with areas of hemorrhage and chorionic villi, which confirmed a right ovarian gestation (Figure 3).

The patient recovered well after surgery and was discharged home 5 days postoperatively.  $\beta$ -HCG concentrations were undetectable 3 weeks after the surgery. The patient and her husband were satisfied with the treatment.

The study received the approval of the ethical committee of the Second Hospital of Hebei Medical University (approval number: 2022-P029). Written consent was obtained from the patient for the use of their clinical information and images used in the case report. We have de-identified the details such that the identity of the patient may not be ascertained in any way. The reporting of this study conforms to the CARE guidelines.<sup>6</sup>



**Figure 3.** Histology shows right ovary tissue. The villi (arrow) are surrounded by ovarian tissue and hemorrhage can be seen.

## Literature review

We performed a literature review using a MedLine and PubMed search with the keywords “ovarian pregnancy”, “primary ovarian ectopic pregnancy”, “*in vitro* fertilization embryo transfer”, “frozen–thawed embryo transfer”, “assisted reproductive technology” “IVF-ET”, “FET”, “ART”, and “bilateral salpingectomy”. We found four cases of OP after IVF-ET with previous bilateral salpingectomy in the literature from 1954 to the present<sup>2–5</sup> (Table 1).

## Discussion

In 1968, some researchers<sup>7</sup> suggested that there are two types (intrafollicular and extrafollicular) of OP, and less is known about the intrafollicular type. Dursun *et al.*<sup>8</sup> stated that, in the primary type of ovarian ectopic pregnancy, the ovum is fertilized in the peritoneal cavity and then it is implanted into the ovary. Secondary ovarian ectopic pregnancy develops as a cause of tubal abortion of the embryo and then it is implanted in the ovary. Four criteria have been proposed to distinguish OP from abnormal pregnancies, and one of them is that the fallopian tube with its fimbria should be intact and separate from the ovary. However, the patient diagnosed with OP in the present study did not have the fallopian tubes. The mechanism of OP after IVF-ET with previous bilateral salpingectomy is unknown.

In 2014, Shan *et al.*<sup>9</sup> showed that pelvic inflammatory disease, previous surgery in the pelvic area, and intrauterine device use may be associated with OP. These factors are generally thought to be associated with ovarian inflammation, which leads to a reduction in follicular fluid pressure and ovulation disorder. The ovum is then trapped in the ruptured follicles and fertilization only occurs in the ovary. Zhu *et al.*<sup>10</sup> found that IVF-ET was a risk factor for

**Table 1.** Review of cases of ovarian pregnancy following in vitro fertilization-embryo transfer with previous bilateral salpingectomy.

Author	Age (y)	Indication	Attempt	Day	Number of transferred embryos	USG follow-up	Serum $\beta$ -HCG (mIU/mL)	Symptoms and signs	Surgery	Pathology	Implantation site	Prognosis
Hsu et al. <sup>2</sup>	29	Bilateral salpingectomy for hydrosalpinx (laparoscopy)	2nd	5th	3	No IU gestation, adnexal mass	2748	Acute abdominal pain	Methotrexate at admission and then laparoscopic enucleation	Ovarian pregnancy	Left ovary	NA
Seshadri et al. <sup>3</sup>	37	Bilateral salpingectomy for an ectopic pregnancy (laparotomy)	3rd	NA	2	Ovarian gestation, corpus luteum	110,149	Lower abdominal pain	Ovarian wedge resection by laparotomy	NA	Right ovary	An uneventful postoperative recovery and gradual decrease in HCG concentrations
Cruciani et al. <sup>4</sup>	26	Bilateral salpingectomy for hydrosalpinx (laparoscopy)	1st	NA	3	No IU gestation, adnexal mass	22,368	Lower abdominal pain	Laparoscopic enucleation	NA	Right ovary	An uneventful postoperative recovery and gradual decrease in HCG concentrations
Feit et al. <sup>5</sup>	29	Bilateral salpingectomy for an ectopic pregnancy (laparoscopy)	1st	NA	NA	No IU gestation, adnexal mass	2366	Lower abdominal pain	Laparoscopic ovarian wedge resection	Ovarian pregnancy	Right ovary	NA

USG, ultrasonography;  $\beta$ -HCG, beta-human chorionic gonadotropin; IU, intra-uterine; NA, not available.

OP. Possible causes of OP include the reverse migration of embryos through the fallopian tubes, the volume of the injected culture medium, a difficult ET, and even perforation of the uterus during ET. These pathological processes may explain why the implantation site occurs in the ovary, but they cannot explain how the embryo ended up in the ovary in this case.

Hsu *et al.*<sup>2</sup> and Seshadri *et al.*<sup>3</sup> suggested that dissecting the fallopian tubes with electrocauterization can cause a reduced blood supply and poor healing at the uterocornual region, which might result in pressure necrosis and then fistula formation after salpingectomy. This situation is likely to have occurred in our case. Feit *et al.*<sup>5</sup> considered that, while transferring the embryo via an ET tube, migration of the embryo through a tuboperitoneal fistula may occur. In our case, the right tubal stump was adherent to the right ovary, and a tract could not be seen by the naked eye, but a cornual fistula was the most likely reason for OP.

In addition, during controlled ovarian hyperstimulation, high drug concentrations cause an increase in steroid hormone concentrations, the ovaries are overstimulated, and the ratio of serum estrogen and progesterone changes. This situation increases the sensitivity of constriction of uterine smooth muscle,<sup>11</sup> and strong endometrial waves in the fundal area of the uterus can move embryos into the cornual fistulae. Oocyte retrieval may lead to the formation of ovarian surface scars<sup>3</sup> and a fistulous communication between them, resulting in ectopic implantation from the uterus to the ovary. However, in this case, ET involved a frozen-thawed cycle rather than a fresh cycle, there was no ovarian stimulation, and the scar may have recovered within several months after oocyte retrieval. However, regardless of using frozen ET, the above-mentioned factors cannot be ruled out as a possible etiology for OP.

At the same time, the patient had a history of open surgery for ectopic pregnancy twice, and extensive adhesion of the pelvis and abdominal cavity was observed during the operation. Additionally, inflammatory factors cannot be completely excluded.

The clinical manifestations of OP are easily confused with gynecological acute abdomen, and the preoperative diagnostic rate is low. The diagnosis of OP is mainly based on intraoperative exploration and postoperative pathological results. Color Doppler ultrasound plays an important role in the preoperative diagnosis of OP. With the progress of three-dimensional technology, three-dimensional ultrasound can be used in medicine to distinguish OP from an ovarian cyst, which is expected to improve the preoperative diagnostic rate of OP.

With the development of laparoscopic technology, laparoscopic conservative surgery is the main treatment of OP.<sup>12</sup> A common surgical procedure for OP is ovarian cystectomy or ovarian wedge resection, which can achieve ovarian preservation. By enucleating the gestational sac bluntly from the ovary and stopping blood by suture, not by bipolar coagulation, the surrounding ovarian tissue is protected to the greatest possible extent in patients suffering from infertility. This method was also used in our case. In a ruptured ectopic pregnancy accompanied by severe bleeding, a partial oophorectomy can be considered.

Besides salpingectomy for a tubal pregnancy, this procedure is the preferred pretreatment for hydrosalpinx before IVF-ET to improve the chance of pregnancy and a live birth.<sup>13,14</sup> Surgeons must pay attention to the details of salpingectomy to avoid a heterotopic pregnancy in patients with IVF. In our case, the stump of the right fallopian tube was a little long with a loose knot on it. The presence of the fallopian tube stump and the formation of a uterine cornual

fistula were the main causes of the ectopic pregnancy. Therefore, to eliminate the risk of an ectopic pregnancy, all fallopian tubes should be removed as completely as possible, even for bilateral fallopian tube resection.<sup>15</sup> Aiding uterine cornual suture by silk is recommended. The interstitial part of the fallopian tubes was sutured, and electrocoagulation was performed at the end of the interstitial part of the fallopian tubes in our case. This suggests that this technique can prevent the formation of a postoperative fistula caused by inadequate electrocoagulation, and thus prevent migration of the embryo out of the uterus.

In conclusion, the mechanism of ovarian pregnancy is unclear. The findings in our case suggest that the formation of a uterine horn fistula after bilateral salpingectomy may promote the occurrence of OP. According to the technical standard of an operation, the interstitial part of the fallopian tubes should be removed close to the uterine angle during the operation, and the tubal stump should be closed by ligation at the proximal end of the operation. Additionally, a silk thread should be used for all ligations. In addition, resection of irreparable tubal diseases may not necessarily achieve positive clinical effects. In patients with double tube resection and high-risk factors for OP, especially those with assisted reproductive technology, clinicians and ultrasound technicians should be vigilant, be familiar with and be proficient in the diagnosis and treatment of OP, avoid misdiagnosis and mistreatment, perform an early diagnosis and early treatment, and maximize the protection of the patient's fertility.

### Acknowledgements

We thank the patient and their family in agreeing to share their experience to provide guidance in treating the rare condition of OP.

### Author contributions

Mingle Zhang contributed to the conception of the study, performed data analysis, and wrote the manuscript. Caijun Zhao, Jiahua Zheng, and Zhongkang Li contributed to preparing the manuscript. Meiyun yin, Haiyan Li, and Yuan Wang contributed to managing the patient. Xianghua Huang helped perform the analysis with constructive discussions.

### Data availability statement

The datasets used during the current study are available from the corresponding author on reasonable request.

### Declaration of conflicting interest

The authors declare that there is no conflict of interest.

### Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

### ORCID iD

Xianghua Huang  <https://orcid.org/0000-0002-4609-1222>

### References

1. Joseph RJ and Irvine LM. Ovarian ectopic pregnancy: Aetiology, diagnosis, and challenges in surgical management. *J Obstet Gynaecol* 2012; 32: 472–474.
2. Hsu CC, Yang TT and Hsu CT. Ovarian pregnancy resulting from cornual fistulae in a woman who had undergone bilateral salpingectomy. *Fertil Steril* 2005; 83: 205–207.
3. Seshadri S, Shirley P, Jaiganesh T, et al. In vitro fertilisation and embryo transfer for bilateral salpingectomies results in a ruptured ovarian ectopic pregnancy due to a tubal stump fistula: a case report and review of the literature. *Case Reports* 2010; 2010: r920092291.
4. Cruciani L, Gerli S, Baiocchi G, et al. Ovarian pregnancy after in vitro fertilisation in a woman with previous bilateral



- salpingectomy. *J Obstet Gynaecol* 2011; 31: 270–271.
5. Feit H, Leibovitz Z, Kerner R, et al. Ovarian Pregnancy Following in Vitro Fertilization in a Woman After Bilateral Salpingectomy: A Case Report and Review of the Literature. *J Minim Invas Gyn* 2015; 22: 675–677.
  6. Gagnier JJ, Kienle G, Altman DG, et al; CARE Group. The CARE guidelines: consensus-based clinical case reporting guideline development. *Headache* 2013; 53: 1541–1547.
  7. Tan KK and Yeo OH. Primary ovarian pregnancy. *Am J Obstet Gynecol* 1968; 100: 240–249.
  8. Dursun P, Gultekin M and Zeyneloglu HB. Ovarian ectopic pregnancy after ICSI-ET: a case report and literature review. *Arch Gynecol Obstet* 2008; 278: 191–193.
  9. Shan N, Deng W, Fu Y, et al. Unusual ectopic pregnancies: a retrospective analysis of 65 cases. *J Obstet Gynaecol Res* 2014; 40: 147–154.
  10. Zhu Q, Li C, Zhao WH, et al. Risk factors and clinical features of ovarian pregnancy: a case-control study. *BMJ Open* 2014; 4: e006447.
  11. Paltieli Y, Eibschitz I, Ziskind G, et al. High progesterone levels and ciliary dysfunction—a possible cause of ectopic pregnancy. *J Assist Reprod Genet* 2000; 17: 103–106.
  12. Scutiero G, Di Gioia P, Spada A, et al. Primary ovarian pregnancy and its management[J]. *JSLs* 2012; 16: 492–494.
  13. Strandell A, Lindhard A, Waldenström U, et al. Hydrosalpinx and IVF outcome: a prospective, randomized multicentre trial in Scandinavia on salpingectomy prior to IVF\*. *Hum Reprod* 1999; 14: 2762–2769.
  14. Chen J, Huang D, Shi L, et al. Cornual Suture at the Time of Laparoscopic Salpingectomy Reduces the Incidence of Interstitial Pregnancy after In Vitro Fertilization. *J Minim Invas Gyn* 2018; 25: 1080–1087.
  15. OuYang Z, Yin Q, Wu J, et al. Ectopic pregnancy following in vitro fertilization after bilateral salpingectomy: A review of the literature. *Eur J Obstet Gyn R B* 2020; 254: 11–14.