Case Report

Analysis of Surgical Procedure of Four Cases of Ovarian Pregnancies Treated with Laparoscopic Surgery

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Abstract

Ovarian pregnancy is a rare disease, accounting for 0.5%–3% of ectopic pregnancies. Ovarian pregnancy risk factors and preoperative diagnosis have been extensively reported. However, its histopathology and surgical findings have been poorly studied. To examine appropriate surgical procedures, we investigated the clinical features, surgical findings, and histopathological examinations of four ovarian pregnancy cases treated in our hospital. In histopathological examination, most specimens did not contain ovarian tissues; in some cases, villous tissues were buried in a clot. Therefore, evaluating the appropriateness of surgical resection range from histopathological images was difficult. However, the postoperative course was favorable; no cases manifested complications. Considering all these facts, we regarded the surgical procedures of the four cases in this study as appropriate. For the treatment of ovarian pregnancies, especially for the outward development type, a sufficient therapeutic effect may be achieved even without extensive excision of the ovarian tissues by laparoscopic surgery.

Keywords: Extrafollicular type, histopathological examinations, laparoscopic surgery, ovarian pregnancy, ovarian reserve

Introduction

Ovarian pregnancy is a rare disease that accounts for 0.5%–3% of ectopic pregnancies.[1] This condition has no specific diagnostic method, and preoperative diagnosis is difficult. Massive intraperitoneal bleeding may occur due to the delayed diagnosis of ovarian pregnancy; hence, immediate blood transfusion is required.[2] Ovarian pregnancy risk factors and preoperative diagnosis have been extensively reported.[3] However, its histopathology and surgical findings have been poorly studied. Hence, we investigated the clinical features, surgical findings, and histopathological examinations of four patients who underwent surgical treatment for ovarian pregnancy at our hospital and explored appropriate surgical procedures.

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Case Reports

Case 1

A 31-year-old female (gravida 2, parity 1) at 7 weeks and 2 days of gestation was admitted to our hospital. Her medical history stated no record of the use of an intrauterine device (IUD), previous chlamydial infection, endometriosis, or pelvic inflammatory disease. Upon examination, there was mild tenderness in all sides of her abdomen; her serum hCG was 3941 mIU/mL. However, the ultrasound revealed no gestational sac in the uterus, thereby strongly suggesting ectopic pregnancy. Hence, laparoscopic operation was performed. Marked intra-abdominal hemorrhage and hematoma in the right ovary were found [Figure 1]. Intraoperatively, she progressed to hemorrhagic shock; thus, blood transfusion was performed. The patient was diagnosed

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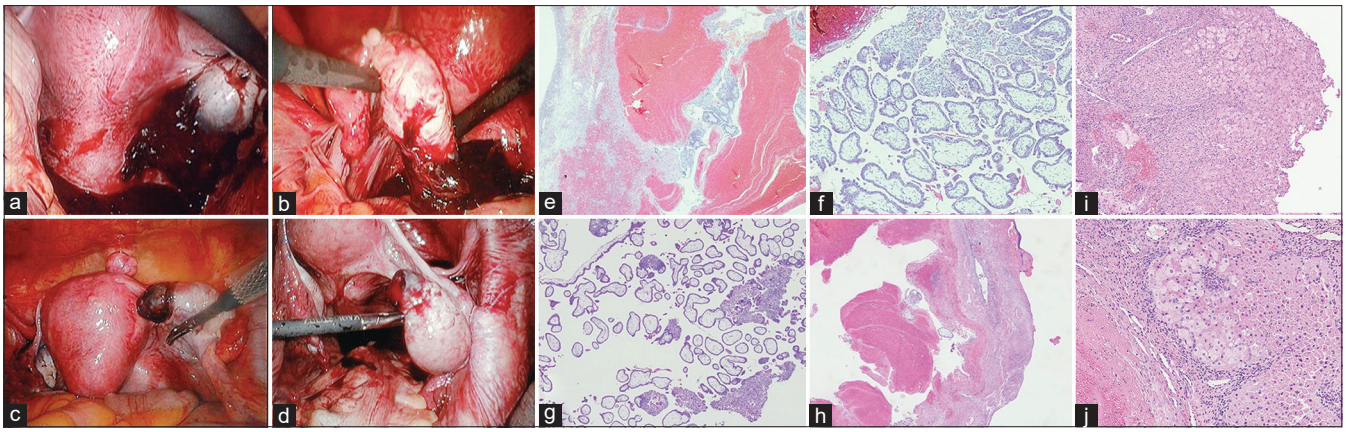

Figure 1: Intraoperative and pathological findings of four patients with ovarian pregnancy. Laparoscopic images of (a-d) correspond to cases 1, 2, 3, and 4, respectively. Pathological images of (e-h) correspond to cases 1, 2, 3, and 4, respectively (low-power field). Pathological images (i and j) illustrate the corpus luteum and villus tissues in cases 3 and 4, respectively (low-power field)

with ovarian pregnancy, and the hematoma in the right ovary was removed using a monopolar electrical scalpel, preserving as much ovarian tissue as possible. The total blood loss was approximately 800 g, and the total operation time was 58 min. Histopathological examination revealed villus tissues buried in the hematoma on the ovarian surface. Ovarian tissues were also noted in the specimen, but its positional relationship with the villus tissues could not be evaluated.

Case 2

A 33-year-old gravida 1, nulliparous female at 5 weeks and 5 days of gestation was admitted in our hospital. She had no risk factors for ectopic pregnancy such as the use of an IUD, previous chlamydial infection, endometriosis, and pelvic inflammatory disease. The patient had unexplained infertility and achieved pregnancy by *in vitro* fertilization and embryo transfer (IVF-ET). On examination, she was hemodynamically stable with a soft abdomen. There was mild tenderness in the left iliac fossa. Despite the high level of serum hCG concentration (5312 mIU/mL), no gestational sac was found in the uterus, possibly indicating ectopic pregnancy. Hence, emergency surgery was performed. We resected the left ovarian mass using monopolar electrocautery and an electrothermal bipolar-activated device. The operation was completed with a small amount of blood loss and an operation time of 94 min [Figure 1]. Histopathological examination revealed the presence of villous tissues at the early pregnancy stage, but no ovarian tissues were found.

Case 3

A 26-year-old gravida 1, nulliparous female was presented to our hospital. Her medical history stated no record of the use of an IUD, endometriosis, or pelvic inflammatory disease. She was at 8 weeks of gestation and asymptomatic. Ultrasound examination could not confirm the gestational sac in the uterus, and her serum hCG was 9865 mIU/mL. Thus, ectopic pregnancy was highly suspected, and emergency laparoscopic surgery was performed. Intraoperatively, a right ovarian pregnancy was identified; hence, the pregnancy tissues were resected using an electrothermal bipolar-activated device, preserving as much ovarian tissue as possible. The total blood loss was 50 g, and the operation time was 73 min [Figure 1]. Histopathological examination revealed villous tissues at the early pregnancy stage, but the specimen was fragmented. Thus, the positional relationship between the villous and ovarian tissues was difficult to evaluate. In addition, a part of the ovarian stroma exhibited hemorrhagic corpus luteum and decidual-like changes.

Case 4

A 32-year-old female (gravida 2, parity 1) was presented to our hospital. Her medical history stated no record of the use of an IUD, previous chlamydial infection, endometriosis, or pelvic inflammatory disease. She was at 8 weeks of gestation, as estimated from the last menstrual period. She was hemodynamically stable but had rebound tenderness on the left iliac fossa. Ultrasound revealed swelling of the right appendage and fluid accumulation in the Douglas fossa, suggesting ectopic pregnancy. An extrovertically growing hematoma was found in the right ovary. An electrothermal bipolar-activated device was used to incise the borderline between the ectopic pregnancy and normal ovarian tissues. The ectopic pregnancy was excised, ensuring ovary conservation. The blood loss was 100 g; the operation time was 77 min.

Histopathological examination revealed degenerated trophoblasts in the ovary. The distance from the ovary surface to the villous tissue was 7 mm. Moreover, no continuity was found between the villus tissue and corpus luteum.

Discussion

This study suggests that performing laparoscopic surgery for ovarian pregnancy with a mass located outside the ovary

may yield a sufficient therapeutic effect without aggressive ovarian resection.

Diagnosis and treatment of ovarian pregnancy continues to be challenging for clinicians because no typical risk factors exist compared with other types of ectopic pregnancy.[2] To examine appropriate surgical procedures, we investigated the clinical features, surgical findings, and histopathological examinations of four ovarian pregnancy cases treated in our hospital [Table 1].

This clinical study was approved by our ethics committee (31-075 [9574]).

The median age of the patients was 31.5 ± 3.1 years. Cases 1 and 2 were infertile and achieved pregnancy by IVF-ET, but none of them had any history of smoking, pelvic peritonitis, or IUD insertion. The gestational age during surgery was 5–8 weeks, and the serum hCG value was 5319 ± 3294 mIU/mL. In all cases, it was difficult to make a differential diagnosis for tubal abortion and ovarian pregnancy. According to surgical findings, intraperitoneal hemorrhage occurred, and a mass, mainly composed of hematoma, developed outside of the ovary. The average operation time was 74 ± 13 min, and the average blood loss during the operation was 242 ± 373 g. All ovarian masses were resected at the borderline with normal ovaries and not deep into the normal ovarian tissue. All the cases could be treated solely with laparoscopic surgery. However, in case 1, blood transfusion was required because of hemorrhagic shock. It was speculated that villous tissue was more likely to be implanted on the ovary surface and rich in angiogenesis than that in the other three cases, resulting in massive intraperitoneal hemorrhage.

Currently, diagnosis is made using the criteria described by Spiegelberg, which includes the fact that the ovary is attached to the uterus by the ovarian ligament, gestational sac is located at the position of the ovary, the fallopian tube is intact with its fimbria and separated from the ovary, and ovarian tissue is present in the specimen histologically.[4] The pathological examination is shown in Figure 1. In case 1, the clot and villus tissue were mixed; thus, measuring the distance between the ovarian tissue and villus tissue was difficult. Case 2 also had villous tissues adjacent to the clot, but no ovarian tissue was identified. In case 3, the specimen itself was fragmented; however, villus tissues were found and not ovarian tissues. In case 4, the distance from the ovary surface to the villous tissue was 7 mm. In all cases, no continuity was observed between the corpus luteum and villus tissue. In most cases, Spiegelberg's criteria cannot be satisfied because most specimens did not contain ovarian tissues, and in some cases, villous tissues were buried in a clot. However, Chelmow *et al*. proposed a revision of the Spiegelberg's criteria for the diagnosis of an ovarian pregnancy to allow laparoscopic diagnosis and medical management.[5] Here, all cases met Chelmow's criteria because the villous tissue was identified in all specimens.

In addition, evaluating the appropriateness of surgical resection range from histopathological images was difficult. However, the postoperative course was favorable, and no cases manifested complications. Considering all these facts, we regarded the surgical procedures of the four cases in this study as appropriate.

Ovarian pregnancy is classified into intrafollicular ovarian pregnancy (intrafollicular type) and extrafollicular ovarian pregnancy (extrafollicular type) from the viewpoint of

Table 1: Demographic characteristics and surgical findings of four patients with ovarian pregnancy

Case No	1	2	3	4
Age (yrs)	31	33	26	32
Gravidity	2	1	1	2
Parity	1	0	0	1
Method for pregnancy	IVF	IVF	-	-
BMI (kg/m2)	18.1	20	21.5	18.4
IUD use	-	-	-	-
Smoking	-	-	-	-
GA (days)	51	40	56	58
hCG (mIU/mL)	3941	5312	9865	2158
Hemoperitoneum (mL)	800	10	50	100
Blood transfusion	+	-	-	-
Laterality	Right	Left	Right	Right
Operation time (min)	58	94	73	77
Ovarian preservation	preserved	preserved	preserved	preserved
Postoperative complication	-	-	-	-
Subsequent pregnancy	unknown	unknown	unknown	unknown

BMI: Body mass index, IVF = In vitro fertilization, IUD = intrauterine contraceptive device, GA = Gestational age

etiology.[6] In the intrafollicular type, the oocyte is not discharged from the follicle during ovulation, and the sperm enters from the ruptured opening and then fertilized in the follicle. The extrafollicular type occurs when an oocyte has been excreted once out of the follicle and implanted on the surface of the ovary after fertilization. In the intrafollicular type, continuity exists between the corpus luteum and villi, but in the extrafollicular type, the developed corpus luteum is found away from the implantation site. In all of the four examined cases, no continuity was detected in the villous tissue and corpus luteum, suggesting an extrafollicular-type ovarian pregnancy.

Moreover, surgical findings have also been classified. Nakagawa *et al*. categorized 13 ovarian pregnancies into outward development type (ten cases) and mass formation type (three cases) according to surgical findings. These two developmental patterns may possibly correspond to the clinical phenotypes of the intrafollicular and extrafollicular types.[7] All of the four examined cases were considered to be outward-growing ovarian pregnancies. Most cases did not contain enough ovarian tissues in the excised specimen. We were concerned that these findings may be due to insufficient excision of the villus tissue. Nonetheless, the postoperative course was favorable in all cases, and the surgical procedure performed this time was considered appropriate.

All cases examined here were outward-growing types; the surgical procedure suitable for the mass-forming type may not be the same. Kaur *et al*. have reported laparoscopic treatment for the mass-forming type of ovarian pregnancy; before mass resection, argipressin was locally injected into the border of the normal ovary and the tumor.[8] Thus, we may need to optimize surgical procedures according to the type of ovarian pregnancy.

Surgical treatment for ovarian pregnancy needs a direct approach to the ovary, but it may be potentially hazardous to the ovarian reserve. Nevertheless, the disease is in the acute phase due to its character. Therefore, reports on postoperative ovarian reserve for ovarian pregnancy are limited. Koo *et al*. reported long-term outcomes of 28 ovarian pregnancies that were followed up for at least 1 year after surgery.[9] No recurrence cases were found, one case (3.6%) had secondary infertility, and 13 cases (46.4%) were finally pregnant. Long-term follow-up is necessary for patients who want to raise children.

In conclusion, for the treatment of ovarian pregnancies, especially for the outward development type, a sufficient therapeutic effect might be achieved even without extensive excision of the ovarian tissues by laparoscopic surgery. However, the ovarian reserve after laparoscopic surgery for ovarian pregnancy remains unknown; hence, long-term follow-up should be considered.

Ethical approval

This study was approved by IRB in The Jikei University School of Medicine (approved number: 30-321(9342)) and the need for informed consent was waived by the IRB. We provided the patients in this research with the opportunity to opt out.

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Conflicts of interest

There are no conflicts of interest.

References

1. Raziel A, Schachter M, Mordechai E, Friedler S, Panski M, Ron-El R. Ovarian pregnancy-a 12-year experience of 19 cases in one institution. Eur J Obstet Gynecol Reprod Biol 2004;114:92-6.
2. Odejinmi F, Rizzuto MI, Macrae R, Olowu O, Hussain M. Diagnosis and laparoscopic management of 12 consecutive cases of ovarian pregnancy and review of literature. J Minim Invasive Gynecol 2009;16:354-9.
3. Seo MR, Choi JS, Bae J, Lee WM, Eom JM, Lee E, *et al*. Preoperative diagnostic clues to ovarian pregnancy: Retrospective chart review of women with ovarian and tubal pregnancy. Obstet Gynecol Sci 2017;60:462-8.
4. Spiegelberg O. Zur Cosuistik der ovarialschwanger schalt. Arch Gynaekol 1973;13:73-6.
5. Chelmow D, Gates E, Penzias AS. Laparoscopic diagnosis and methotrexate treatment of an ovarian pregnancy: A case report. Fertil Steril 1994;62:879-81.
6. Tan KK, Yeo OH. Primary ovarian pregnancy. Am J Obstet Gynecol 1968;100:240-9.
7. Nakagawa K, Enari T, Kaneko E, Kawamura H, Santou A, Uesato T, *et al*. Ovarian pregnancy: Report of 13 cases in recent ten years. Jpn J Gynecol Obstet Endosc 2004;20:158-62.
8. Kaur N, Reid F, Ma K. Ovarian ectopic pregnancy: Laparoscopic excision and ovarian conservation. J Minim Invasive Gynecol 2019;26:1006.
9. Koo YJ, Choi HJ, Im KS, Jung HJ, Kwon YS. Pregnancy outcomes after surgical treatment of ovarian pregnancy. Int J Gynaecol Obstet 2011;114:97-100.