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Successful management of heterotopic cornual pregnancy with laparoscopic cornual resection



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ABSTRACT

Objective: To examine the feasibility of laparoscopic cornual resection for the treatment of heterotopic cornual pregnancy.

Study design: Women who underwent laparoscopic cornual resection for heterotopic cornual pregnancy at our hospital between January 2003 and March 2015 were retrospectively analyzed. We evaluated significant parameters such as operative complications and postoperative pregnancy outcomes of concomitant pregnancy.

Results: Thirteen patients with heterotopic cornual pregnancy were included in the study. All were pregnant through assisted reproductive technology, and the diagnosis was made at a median of 6 + 6 weeks (range 5 + 4–10 + 0). They were successfully treated with laparoscopic cornual resection and admitted for a median of 4 days (range, 2–7) postoperatively. The median operative time was 65 min (range, 35–145 min) and estimated blood loss was 200 mL (range, 10–3000 mL). There was a spontaneous abortion at 7 + 6 gestational weeks in a patient who received bilateral cornual resection. Seven patients delivered babies at term and 3 at preterm. All 10 women delivered without any maternal or neonatal complications. Two were lost to follow-up.

Conclusions: Laparoscopic cornual resection is a feasible primary approach for the management of heterotopic cornual pregnancy.

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Introduction

Heterotopic pregnancy is defined as the coexistence of an intrauterine pregnancy and an extrauterine pregnancy. Heterotopic pregnancy occurs at various sites, and may present as bilateral tubal pregnancy, abdominal and intrauterine pregnancy, twin tubal and intrauterine pregnancy, and simultaneous intrauterine and cornual pregnancies [1]. The reported incidence of heterotopic pregnancy in a spontaneous cycle is 1 in 30,000 [2]. However, with increasing use of assisted reproductive technology (ART) procedures such as in vitro fertilization (IVF) and embryo transfer (ET), the incidence of heterotopic pregnancy

has increased to 1 in 100 pregnancies with ART [3]. Cornual pregnancy occurs when the embryo implants in the intramural portion of the Fallopian tube. It is rare, accounting for around 2% of ectopic pregnancies [4]. The incidence of heterotopic cornual pregnancy after an IVF cycle is not known. However, the calculated incidence from data based on the 1995 reproductive health report is estimated to be as high as 1:3600 IVF pregnancies [1].

There are several options for the treatment of heterotopic cornual pregnancy; surgical, medical or expectant treatment. The patient can be treated surgically by cornual resection or hysterectomy either by laparotomy or laparoscopy. Another option is the direct injection of potassium chloride (KCl), hypertonic solution, and methotrexate into the ectopic gestational sac [1–3,5]. Lastly, if the patient has no symptoms and fetal death in the cornual gestation is confirmed by ultrasonography, expectant management can be used [6].

We have adopted laparoscopic cornual resection as the first-line approach for the treatment of this rare but serious condition. This study was performed to demonstrate the feasibility of

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laparoscopic cornual resection in treating heterotopic cornual pregnancy and to examine the obstetric outcome of an accompanying pregnancy after surgery.

Materials and methods

The study was approved by the institutional review board (IRB) of CHA Gangnam Medical Center (GCI-15-13). After obtaining IRB approval, the authors retrospectively reviewed the medical records of the patients with diagnosed heterotopic cornual pregnancy between January 2003 and March 2015.

The patient data included age, parity, gestational weeks at diagnosis, hemoglobin (Hb) level on the pre- and postoperative day, length of hospital stay, operative time, and obstetric outcomes. The diagnosis of heterotopic cornual pregnancy was established by ultrasonographic findings. In our institution, the diagnosis of heterotopic cornual pregnancy was made by transvaginal ultrasonography using the following criteria: intrauterine pregnancy complicated by cornual ectopic pregnancy, i.e. a gestational sac visualized high in the fundus, and not surrounded by 5 mm of myometrium in all planes; and a gestational sac seen separately and < 1 cm from the most lateral edge of the uterine cavity [1,7,8].

Laparoscopic procedures were as follows. The patient was placed in dorso-lithotomy position. General endotracheal anesthesia was initiated after the preparations for surgery were completed. A 12-mm trocar was directly inserted at the umbilicus through a vertical skin incision. The abdomen was insufflated with CO₂. After cornual pregnancy was confirmed with a 5-mm endoscope, three additional 5-mm ancillary trocars were placed in the left lower quadrant, right lower quadrant, and suprapubic area, respectively. The intra-abdominal pressure was maintained at 12 mmHg during surgery. A circumferential incision was made with monopolar hook or harmonic shears around the cornual pregnancy. The patient underwent laparoscopic cornual resection either with or without ipsilateral salpingectomy according to the surgeon's decision. We tried to secure hemostasis with Vicryl 1-0

suture. Hemostasis with bipolar cautery was minimized to reduce thermal damage to the viable intrauterine gestation. The uterine defect was repaired with either single or double layers using interrupted suture technique with Vicryl 1-0 and Vicryl 2-0 (Fig. 1). The resection materials were removed by using a laparoscopic retrieval bag.

After the operation, fetal heart beat was checked with Doppler in the recovery room. The patient was observed for at least 24 h. On postoperative day 1, transvaginal ultrasonography was performed to confirm fetal viability.

Results

From January 2003 through January 2015, a total 3400 cases of ectopic pregnancy were diagnosed in our institution. Among them, 97 (2.9%) were diagnosed with a cornual pregnancy. There were 80 cases (2.4%) of heterotopic pregnancy. A total of 13 cases (0.4%) of heterotopic cornual pregnancy were diagnosed in our hospital.

Table 1 shows the patient characteristics and surgery outcomes. The median age and parity at the time of admission were 31 years (range, 25–36) and 0 (range, 0–1), respectively. Three women had at least one previous extrauterine pregnancy history (case nos. 3, 4, 11 in Table 2), among them one patient had experienced 3 times of ectopic pregnancies (case no. 4). All patients had at least one previous pelvic surgery history such as myomectomy, salpingectomy, or oophorectomy and 12 patients had undergone tubal surgery or had tubal pathology. Eight patients experienced salpingectomy previously. Among them, one patient had undergone laparoscopic surgery 4 times: consecutive two times of unilateral salpingectomies due to ectopic pregnancies, myomectomy, and ovarian cystectomy, respectively (case no.4). Including this patient, all 13 women achieved present pregnancies by ART procedure. A median 3 embryos (range, 2–5) were transferred in each patient among whom 4 patients underwent transfer of 4–5 embryos (case nos. 1–4).

The clinical manifestations of the patients were abdominal pain (4 of 13 patients, 30.8%) and vaginal spotting (2 of 13, 15.4%), but

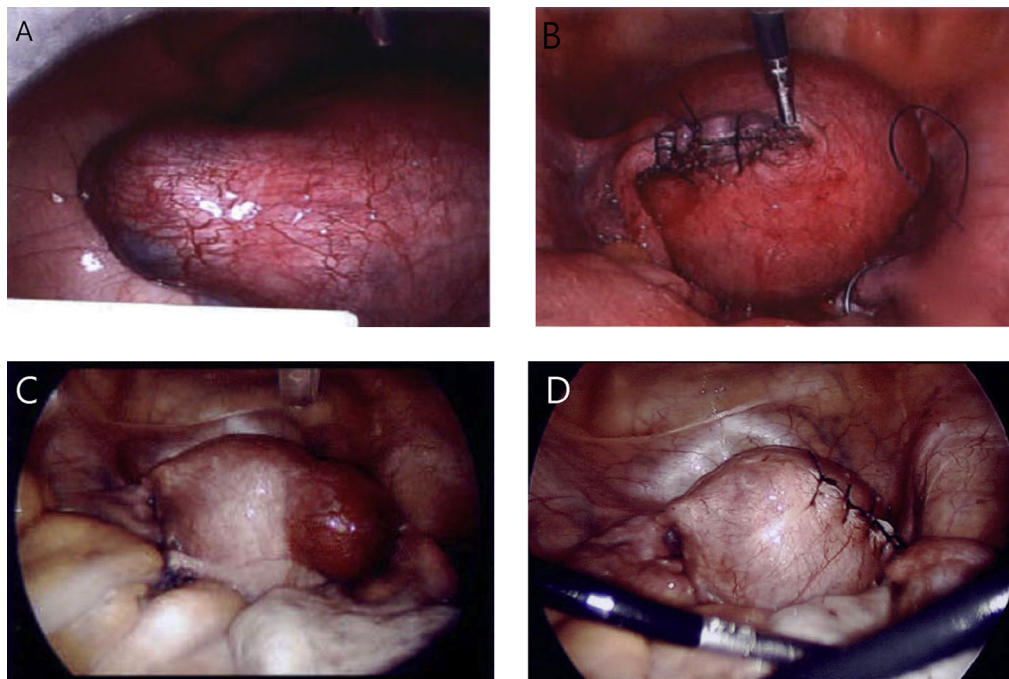


Fig. 1. Pre- and postoperative images of heterotopic cornual pregnancy. (A) Preoperative image of case no. 10. (B) Postoperative image of case no. 10. (C) Preoperative image of case no. 6. (D) Postoperative image of case no. 6.

Table 1
Patient demographics and surgical outcomes (n = 13).

Variable	Median (range)
Age, years	31 (25–36)
Gravidity	1 (1–6)
Parity	0 (0–1)
Gestational age at diagnosis, week + days	6 + 6 (5 + 4–10 + 0)
Embryo heart beat at ectopic site, No. (%)	2 (16.7)
Potential predisposing factors	
In vitro fertilization, no. (%)	13 (100)
No. of embryos transferred	3 (2–5)
Previous extrauterine pregnancy, no. (%)	3 (23.1) ^a
Previous pelvic surgery, no. (%)	13 (100)
Previous tubal surgery, no. (%)	12 (92.3)
Salpingectomy, no. (%)	8 (58.3) ^b
Tubal ligation, no. (%)	2 (16.7)
Tuboplasty, no. (%)	2 (16.7) ^b
Previous tubal reanastomosis, no. (%)	1 (8.3)
Previous tubal mass removal, no. (%)	1 (8.3)
Clinical manifestation	
Abdominal pain, no. (%)	4 (30.8)
Vaginal spotting, no. (%)	2 (15.4)
Incidental diagnosis on ultrasonography, no. (%)	7 (53.8)
Surgical outcome	
Operation time, min	65 (35–145)
Estimated blood loss, mL	200 (10–3000)
Ruptured type, no. (%)	3 (25.0)
Hemoperitoneum, no. (%)	3 (25.0)
Blood transfusion, no. (%)	3 (25.0)
Hb at admission, g/dL	12.7 (7.7–13.6)
Hb on postoperative day 1, g/dL	10.8 (8.2–11.7)
Length of hospital stay postoperatively, day	4 (2–7)

Hb = hemoglobin.

^a One patient had experienced 3 times of ectopic pregnancies (case no. 4 in Table 2).^b Two patients had undergone unilateral salpingectomy and contralateral tuboplasty (case nos. 10 and 13 in Table 2).

7 of all 13 patients (53.8%) were diagnosed with heterotopic cornual pregnancy incidentally on ultrasound examination. The median gestational age at diagnosis was 6 weeks and 6 days (range, 5 + 4–10 + 0 weeks).

All 13 patients with heterotopic cornual pregnancy underwent laparoscopic cornual resection. Ten (76.9%) underwent surgery before rupture of the extrauterine pregnancy. Two (15.4%) were diagnosed with suspicion of ruptured cornual pregnancy, and there was massive hemoperitoneum in the pelvic cavity (1800 and 3000 mL of blood in case nos. 11 and 12, respectively). One patient (7.7%) was diagnosed with heterotopic cornual pregnancy before rupture of the extrauterine pregnancy. However, the cornual pregnancy was ruptured after admission and emergent laparoscopic surgery was performed. This patient also had massive hemoperitoneum (1500 mL in case no. 13).

Twelve patients underwent laparoscopic unilateral cornual resection and one patient (case no. 3) underwent bilateral cornual resection because she had heterotopic triplet pregnancy including bilateral cornual pregnancy. The median operative time was 65 min (range, 35–145 min) and estimated blood loss was 200 mL (range, 10–3000 mL). The median Hb levels on admission and postoperative day 1 were 12.7 g/dL (range, 7.7–13.6 g/dL) and 10.8 g/dL (range, 8.2–11.7 g/dL), respectively. The median length of postoperative hospital stay was 4 days (range, 2–7) without any intraoperative or postoperative complications.

Among all patients, there was one missed abortion 5 days after surgery in the patient undergone bilateral cornual resection (case no. 3). All of the other 12 patients were confirmed to have ongoing pregnancies postoperatively through 13 weeks of gestational age, but 2 patients were lost to follow-up. Six women delivered by cesarean section (4 at term and 2 at preterm); 4 women delivered vaginally (3 at term and 1 at preterm). All their 11 infants were healthy. Among them, 1 preterm baby received care in the neonatal

intensive care unit (NICU) for 1 month, but had no other neonatal complication (case no. 9).

Among 6 women who tried subsequent pregnancy, except 1 patient (case no. 11), 4 women delivered healthy live babies and 1 woman has an ongoing pregnancy through 35 weeks of gestation. The only patient who had a missed abortion postoperatively also included in this subsequent delivery group without any complication (case no. 3).

Comments

There is no consensus for the treatment of heterotopic cornual pregnancy because of its rarity. Therefore, we investigated the pregnancy outcomes and feasibility of laparoscopic surgery for the management of heterotopic cornual pregnancy. Our results demonstrate that laparoscopic cornual resection is a feasible approach to treat heterotopic cornual pregnancy with favorable surgical and obstetric outcomes.

The uterine cornu has abundant vascularity through the branches of the uterine and ovarian arteries. Cornual pregnancy combined with intrauterine pregnancy often results in life-threatening bleeding due to rupture of cornual pregnancy and subsequent fetal loss because of delayed diagnosis [9]. The patient may have no specific symptoms such as abdominal pain or vaginal bleeding. Because of the presence of an intrauterine and an extrauterine gestational sac, serum β -hCG can be normal or even higher and the diagnosis of heterotopic cornual pregnancy is difficult and often delayed [10]. In addition clinicians might ignore the possibility of extrauterine pregnancy in the case of a confirmed intrauterine gestation. Therefore, meticulous care is required in a normal pregnancy after ART procedures if there are risk factors for ectopic pregnancies such as previous history of ectopic pregnancy, pelvic surgery, or pelvic inflammatory disease.

In our study, 7 patients were diagnosed incidentally by ultrasonography without any symptoms as early as 5 + 4 weeks and even as late as 10 weeks. All 13 patients underwent pelvic surgery previously and 12 patients had undergone tubal surgery or had tubal pathology. All conceived through ART and a median 3 embryos were transferred. Such factors are associated with heterotopic cornual pregnancy [1,11].

Thus, clinicians must be aware of the possibility of heterotopic pregnancy in a patient with a previous history of tubal problems and an ART procedure, with the necessity for early serial ultrasonographic monitoring. Moreover, the number of transferred embryos in ART should be controlled to prevent heterotopic pregnancies in patients with risk factors of ectopic pregnancy.

There are several treatment options for heterotopic cornual pregnancy. In the absence of fetal cardiac activity, with early diagnosis in the first trimester, a symptom-free patient, and limited craniocaudal length and gestational sac size, expectant management can be an option [1,12]. This management avoids the complications of medical treatment such as reduction by spontaneous abortion. Nevertheless, if a fetus in the uterine horn is living, this approach cannot be applied. Because of the possibility of continued growth in the cornual sac and rupture, repeated ultrasonography and close monitoring of symptoms are necessary.

If a patient diagnosed with heterotopic cornual pregnancy is hemodynamically stable, nonsurgical treatment may be suggested. The nonsurgical approach consists of injecting KCl or hypertonic solution directly into the ectopic gestational sac under the guidance of transvaginal ultrasonography [2–4]. The main advantage of nonsurgical treatment is that it is less invasive and inexpensive, with quick recovery, minimal blood loss, and a short length of hospital stay compared to surgical treatment. However, nonsurgical treatment has several concerns for cornual rupture associated with ultrasound-guided cornual puncture and subsequent rupture of an

Table 2
Pregnancy outcomes of the patient who underwent laparoscopic cornual resection (n = 13).

No.	Age (years)	Gravidity /parity	Risk factors	No. of embryos transferred	Symptoms	Gestational age at diagnosis (week + days)	Gestational age at surgery	Type of surgery	Operation time (min)	EBL (mL)	Transfusion (units of packed RBC)	Pregnancy outcome	Subsequent pregnancy outcome
1	36	2/0	IVF-ET, LSC-BTL	5		5+4	5+4	LSC- left cornual resection	40	200	0	Follow up loss after 13 weeks of ongoing pregnancy	
2	30	2/1	IVF-ET, Explo-RSO	4		7+4	7+4	LSC-right cornual resection	65	400	0	Follow up loss after 16+5 weeks of ongoing pregnancy	
3	31	2/0	IVF-ET, Explo-LS	4		6+6 ^c	7+1	LSC-bilateral cornual resection ^b	95	400	0	Missed abortion, 5 days after surgery	IVF-twin pregnancy, elective cesarean delivery at 38 weeks
4	34	6/0	IVF-ET, LSC-BS	4	Abdominal pain	6+6	7+0	LSC-right cornual resection	50	100	0	Elective cesarean delivery at 38+4 weeks, 3060 g	
5	25	1/0	IVF-ET, LSC-Right tubal reanastomosis	3	Vaginal bleeding	6+2	6+2	LSC-right cornual resection	70	10	0	VD at 35+6 weeks, 2210 g, McDonald operation	IVF-twin pregnancy, elective cesarean delivery at 35+6 weeks, s/p McDonald surgery
6	35	1/0	IVF-ET, LSC-BS	2	Abdominal pain	6+2	6+5	LSC-right cornual resection	40	10	0	VD at 38+4 weeks, 2920 g	
7	31	1/0	IVF-ET, LSC-BTL	3		6+6 ^c	6+6	LSC-left cornual resection	35	100	0	VD at 39+1 weeks, 2920 g	IVF pregnancy, VD at 38 weeks
8	31	1/0	IVF-ET, LSC-ovarian cystectomy	2		7+1	7+3	LSC-right cornual resection	55	100	0	Elective cesarean delivery at 38+2 weeks, 2600 g	
9	32	1/0	IVF-ET, LSC-Rt adnexal mass removal, BTO	2		6+4	6+5	LSC-left cornual resection	58	50	0	Cesarean delivery at 31+1 weeks, due to PPROM, 1660 g ^d	
10	26	1/0	IVF-ET, LSC-RS ^c Lt tuboplasty	2		10+0	10+0	LSC-left cornual resection	80	1000	0	Cesarean delivery at 35+6 weeks, due to preterm labor, 2680 g, McDonald operation	
11	30	2/0	IVF-ET, LSC-RS	2	Abdominal pain	6+4	6+4	LSC-right cornual resection	145	1800 ^a	0	Cesarean delivery at 39+0 weeks, 3160 g	
12	27	1/0	IVF-ET, LSC-BS, Tuberculosis peritonitis	3	Abdominal pain	6+4	6+4	LSC-left cornual resection	75	3000 ^a	3	VD at 38+5weeks, 2700 g	IVF pregnancy, VD at 40 weeks
13	29	1/0	IVF-ET, LSC-LS ^c Rt tuboplasty	2	Vaginal bleeding	7+3	7+4	LSC-left cornual resection	80	1500 ^a	2	Elective cesarean delivery at 38+0weeks, 3500 g	IVF pregnancy, early abortion → IVF pregnancy again; 35 weeks of ongoing pregnancy

EBL = estimated blood loss; IVF-ET = in vitro fertilization and embryo transfer; Explo-LS = exploratory left salpingectomy; LSC-BS = laparoscopic bilateral salpingectomy; Explo-RSO = exploratory right salpingo-oohprectomy; LSC-RS = laparoscopic right salpingectomy; LSC-LS = laparoscopic left salpingectomy; LSC-BTL = laparoscopic bilateral tubal ligation; BTO = bilateral tubal obstruction; VD = vaginal delivery; PPROM = premature rupture of membrane.

^a Ruptured cornual pregnancies with hemoperitoneum.

^b This patient had bilateral cornual pregnancies.

^c Embryo heart beat on ectopic site.

^d Neonatal intensive care unit (NICU) admission for 1 month due to preterm delivery, without complication.

ectopic pregnancy and massive bleeding [10]. Drugs such as KCl and NaCl that are used for selective reduction in multifetal pregnancy are harmful to a fetus. A drug administered into the cornual gestation sac may diffuse through the myometrium to the intrauterine pregnancy and into the maternal body. KCl has reportedly been associated with intrauterine damage, periventricular leukomalacia, and limb anomalies [13–16]. In addition, the abortion rate in patients who were treated medically was higher than that of those who underwent surgery (50% vs. 13%) [1]. Another concern about conservative treatment is the recurrence of ectopic pregnancy, particularly repeat ipsilateral cornual pregnancy [8].

The surgical approach is the definitive removal of the cornual ectopic mass either by laparoscopy or laparotomy. Grobman et al. recommended cornual excision in larger gestations (> 4 cm) [17]. In the case of rupture of a heterotopic cornual pregnancy, surgery is the only treatment option. Habana et al. reviewed and compared the treatment results of heterotopic cornual pregnancy according to the mode of treatment [1]. In the group receiving surgical treatment, a higher live birth rate and a lower abortion rate were reported than in the group receiving nonsurgical management (60.9% vs. 50% and 13% vs. 50%, respectively). In less than a decade, laparoscopic surgery has been adopted for the treatment of cornual pregnancy [18,19]. The advantages of laparoscopic management over exploratory laparotomy are the shorter hospital stay, fewer surgical wounds, and reduced use of antibiotics and analgesics.

Several authors reported cases of successful laparoscopic removal of unruptured [20–22] or ruptured cornual resection [4] followed by delivery of healthy babies. In our study, we successfully performed laparoscopic cornual resection without intra- and postoperative complications in all 13 (10 unruptured and 3 ruptured) cornual pregnancies. The pregnancy outcome that was confirmed after laparoscopic surgery in this study was promising. Nine patients in this study delivered healthy babies without complications. One patient delivered at 31 gestational weeks, and the baby was admitted to the NICU for 1 month afterwards without neonatal complications. The pregnancy outcome of other 2 patients is unknown, but they were confirmed to have an ongoing pregnancy through 13 gestational weeks, which means there was no detrimental effect in these pregnancies for 8–9 weeks after the surgery. One patient had a missed abortion 5 days after laparoscopic bilateral cornual resection because of an ectopic pregnancy complicated by bilateral cornual pregnancies. It is possible that the abortion was related to more aggressive laparoscopic surgery.

If surgical management fails, a hemostatic hysterectomy resulting in the loss of the patient's fertility and the intrauterine pregnancy may be required. Accordingly, surgeons need to be experienced and skillful in this operative procedure to minimize bleeding events. In addition surgical resection of the uterine cornu also can lead to subsequent uterine rupture during pregnancy. Although there was no uterine rupture during concomitant or subsequent pregnancy in our study, clinicians must perform careful obstetric monitoring postoperatively, and elective cesarean section is usually preferred to vaginal delivery [8].

Recently, several suture techniques to reduce operative hemorrhage have been introduced, including purse-string suture, square suture, encircling suture, endoloop, and double-impact devascularization technique [23,24]. These techniques might be adopted to reduce surgical complication and improve pregnancy outcomes in treating heterotopic cornual pregnancy.

We performed this study to investigate pregnancy outcomes and the feasibility of laparoscopic surgery for patients with

heterotopic cornual pregnancy. There are several limitations. This study was performed retrospectively with a small number of cases. However, heterotopic cornual pregnancy is a rare condition, and there are few data and a limited number of reports in the literature.

This study demonstrated the feasibility of laparoscopic surgery for treatment of heterotopic cornual pregnancy. We suggest laparoscopic cornual resection as the primary treatment for patients with heterotopic cornual pregnancy. Further studies with a prospective design involving multiple institutions are required to confirm the conclusions of this study.

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