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Transvaginal Cornual Pregnancy Reduction Reduces the Risk of Bleeding in Cornual Heterotopic Pregnancy with Severe Pelvic Adhesions: Case Reports and Literature Review

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Cornual Heterotopic Pregnancy; Transvaginal Embryo Reduction; Laparoscopic Cornual Resection

1. Abstract

Objective: The objective of this research is to explore the individualized treatment of Cornual heterotopic pregnancy (CHP).

Methods: This paper describes the treatment and pregnancy outcome of both two patients with CHP. One of these patients underwent transvaginal cornual pregnancy reduction, and the other underwent cornual pregnancy resection. This study reviewed all published cases of CHP.

Results: A 33-year-old woman with a history of previous cesarean section and tubal surgery was found to have a combined intrauterine pregnancy 40 days after frozen embryo transfer. The patient underwent a cornual pregnancy resection with 1500 ml of intraoperative bleeding during the separation of the horn adhesions, the intrauterine fetus survived and a healthy fetus was delivered by cesarean section at 34 weeks of gestation. The other 33-year-old patient with CHP who was found to have an intrauterine combined cornual pregnancy 30 days after ovulation promotion underwent cornual pregnancy reduction and delivered a live fetus at 39 weeks of gestation. There were 41 cases in the published literature, reporting on cornual heterotopic pregnancy. Live birth rates were comparable between the two groups.

Conclusions: Transvaginal cornual pregnancy puncture and laparoscopic cornual resection are the same effective for the treatment of cornual heterotopic pregnancy. For patients with severe pelvic adhesions, cornual pregnancy reduction may be considered to reduce the risk of bleeding.

2. Introduction

Cornual ectopic pregnancies are a type of ectopic pregnancy in which the embryo is implanted at the junction of the uterus and opening of the fallopian tube, on the medial side of the round ligament. There are two types of cornual ectopic pregnancy [1]. The type I gestational sac grows mostly inside the uterine cavity, with a low risk of rupture of the myometrium and insignificant horn protrusion. The type II gestational sac grows mainly outside of the horn of the uterus, with a significant horn protrusion and a high risk of rupture of the myometrium and hemorrhage. A cornual ectopic pregnancy is one of the most life-threatening types of ectopic gestations, with a mortality rate six to seven times higher than that of ectopic pregnancies in general [2]. Heterotopic pregnancies refer to the coexistence of two or more implantation locations [3]. With the use of assisted reproduction techniques including controlled ovarian hyperstimulation and timed intercourse, intrauterine insemination and in vitro fertilization, the incidence has increased from 1/100 to 1/360 [4]. Cornual heterotopic pregnancy (CHP) is the simultaneous existence of intrauterine pregnancy and cornual ectopic pregnancy. Cornual ectopic pregnancies are often considered tricky, and the situation becomes more complex for cornual heterotopic pregnancies. Until now, there has been no consensus on the ideal treatment options for cornual heterotopic pregnancy

[5], When dealing with CHP, it is important not only to safely remove the cornual gestational sac while ensuring the growth of the intrauterine gestational sac, but also to take into account the safety of the treatment and the damage caused to the patient.

The study explores the treatment options and outcome of cornual heterotopic pregnancies through case analysis and literature review. The objective of this research is to explore the individualized treatment of cornual heterotopic pregnancy.

This study was approved by the Ethics Committee of Hangzhou Women's Hospital. And the two patients had provided informed consent for the publication of the cases.

3. Case Presentation

A 33-year-old woman with secondary infertility for two years underwent Assisted Reproductive technology (ART) in Hangzhou Women's Hospital due to tubal factors. In 2016, the patient had a cesarean section due to dystocia. In 2018 she was diagnosed with an ectopic pregnancy and underwent emergency laparoscopic surgery. During the operation, the patient's left fallopian tube was enlarged and thickened by 8cm*5cm*4cm with blood accumulation, and her right fallopian tube was 7cm*8cm*4cm. Intraoperatively, both fallopian tubes were removed. She was diagnosed with a miscarriage type of pregnancy in the ampulla of the left lateral tubal and hydrosalpinx on the right. The patient has diminished ovarian reserve with a blood AMH (Antimullerian hormone） level of 0.99ng/ml. In her first IVF-ET (in vitro fertilization embryo transfer) cycle, she obtained four eggs, two were routinely fertilized, and two valid embryos were frozen. The two embryos were transferred in two successive FET（frozen embryo transfer） cycles. However, the patient did not get pregnant twice. In the Second IVF cycle, two embryos were obtained and transferred. 11 days after her transplant, the patient was tested pregnant with an HCG level of "288.5 mIU/mL". 26 days after her transplant, an ultrasound of twin pregnancy was performed at the local hospital. 40 days after her transplant, ultrasound was performed with a diagnosis of "cornual heterotopic pregnancy, live fetus in right cornual" at the hospital. The ultrasonography found the following: "An intrauterine gestational sac-like echogenicity of approximately 4.4*3.5*3.0 cm in size is seen, and an embryonic echogenicity of 1.7 cm in parietal-rump diameter is seen inside, a primitive heartbeat can be detected; an inhomogeneous echogenicity of approximately 4.2*4.1*3.8 cm in size is seen on the right side of the uterus immediately adjacent to the right uterine horn, and an anechoic echogenicity of approximately 3.4*2.6*2.4 cm in size is seen inside (Figure 1). An echogenic dark area with a parietal-rump diameter of about 1.6 cm is seen within it, and a primitive heartbeat could be detected. This patient had a clear diagnosis of cornual heterotopic pregnancy (Type 1), and an emergency laparoscopic surgery was performed after communication with the patient. Intraoperatively, the right fallopian tube was seen to be absent, and the right uterine horn was protruding outward with a mass of approximately 4*4*3 cm, located lateral to the round ligament, with a congested surface and a large omentum covering it in dense adhesions, with a large number of old blood clots visible on the surface (Figure 2). The mass was densely adherent to the intrinsic ligament of the right ovary. The right omentum and right uterine horn adhesions were separated by bipolar electrocoagulation, the right uterine horn ruptured during the separation process, and chorionic villi were seen to leap out of the rupture. The operation was difficult with intraoperative bleeding of 1500 ml and intraoperative blood transfusion of 2 U. An ultrasound of the abdomen five days after surgery suggested early intrauterine pregnancy with a 2.1 cm long parietal-rump diameter and a detectable primitive heartbeat. The pregnancy was closely monitored. At 34 weeks of gestation, the patient underwent an emergency cesarean section for abdominal pain to prevent uterine rupture, and a live baby was obtained (Table 1).

The other case of CHP was a 33-year-old patient with polycystic ovary syndrome. The patient was treated with ovulation promotion for ovulation disorder. The blood HCG (Human chorionic gonadotropin) was 61.44 mIU/ml nine days after ovulation. 30 days after ovulation, ultrasound showed coronal heterotopic pregnancy. The ultrasound suggested the followings: "Two gestational sacs are visible in the uterine cavity with an echogenic size of about 2.2*1.6*1.3 cm (intrauterine gestational sac) and 1.5*1.4*0.9 cm (right cornual gestational sac, protruding outward and not clearly connected to the uterine cavity). Both yolk sac and germ were visible inside, with bud lengths of about 0.7 cm and 0.3 cm, respectively, and both were visible with primitive heartbeats." This woman delivered a girl three years ago with no previous surgical history and adamantly refused laparoscopic surgery. The patient was treated with cornual pregnancy reduction under transvaginal ultrasound guidance. On a postoperative day, the ultrasound showed that the heartbeat of the intrauterine gestational sac was visible and that the heartbeat of the ectopic horn gestational sac disappeared. The size of the mixed echogenic mass was 2.2*2.2*1.8 cm. The patient had no significant abdominal pain or vaginal bleeding after the operation. On the first day after surgery, the heartbeat of the intrauterine gestational sac was visible on the follow-up examination, and the mixed echogenic mass was 2.6*2.4*1.8cm. The patient was discharged three days after surgery. Ultrasound at 10 weeks of gestation suggests early intrauterine pregnancy with an ectopic mixed echogenic light cluster of 4.2*2.4*2.4 cm. A cesarean section was performed at 39 weeks of gestation due to umbilical cord wrapping. A healthy male infant was born, with a birth weight of 3700g and Apgar 10/10.

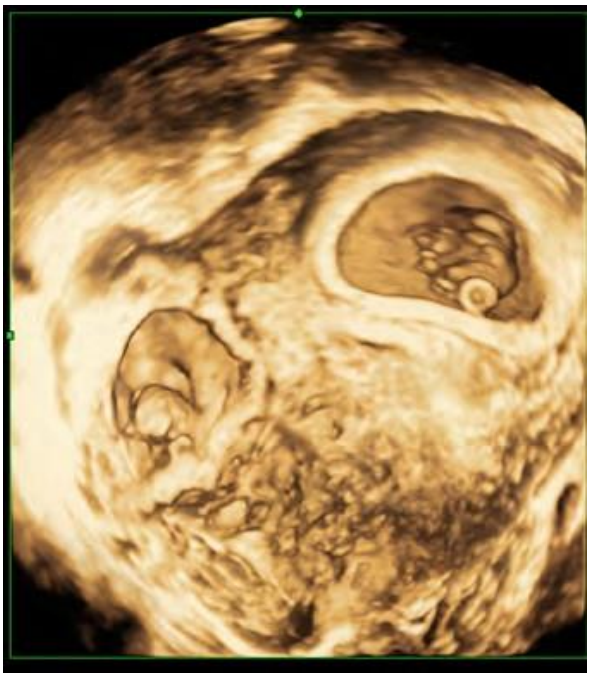

Figure 1: Transvaginal ultrasound showing intrauterine gestational sac and right cornual gestational sac.

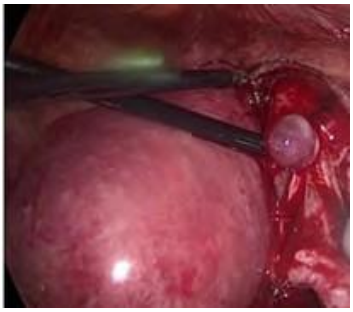

Figure 2: Intraoperative image showing the uterus with a ruptured right cornual pregnancy during the separation of adhesions.

Table 1: 41 cases in the published literature, reporting on cornual heterotopic pregnancy.

Case	Management	Outcome	Authors
10	Cornual Pregnancy Reduction	Cornual rupture 1 Miscarriage 1 Live birth 8	Na E D , Jung I , Choi D H , et al.
1	Diagnostic Laparoscopy	live birth 1	Samuilis A .
1	Laparoscopy Followed by Methotrexate Injection	No miscarriage	Poujade O , Ducarme G , Luton D .
1	Emergency Laparotomy	Miscarriage 1	Cicerone T , Cristina G , Roxana B , et al.
14	Laparoscopic Cornual Pregnancy Resection or Cornual Repair	Cornual rupture 4 Miscarriage 0 Live birth 14	Xu W , Lin X , Huang D , et al.
14	Cornual Pregnancy Reduction (6)	Cornual rupture 2 Miscarriage 1 Live birth 5	Li S , Cao M , Liu H , et al.
	Laparoscopic Cornual Pregnancy Resection (8)	Cornual rupture 0 Miscarriage 1 Live birth 7	

4. Methods

An extensive review of CHP cases from MEDLINE (PUBMED) published was conducted using the search terms ‘‘cornual heterotopic pregnancy’’,” heterotopic pregnancy” and “angular heterotopic pregnancy”. The search was completed by manually searching for references of reports and review articles for relevant case reports. All case reports of heterotopic cornual pregnancies were selected, regardless of management or outcome. Reported data were reported regarding clinical presentation, management and outcomes.

5. Results

41 cases of cornual heterotopic pregnancy were found in published literature. and this study’s cases were not included in the analysis. A total of 16 patients were treated with transvaginal pregnancy reduction and three out of 16 had a cornual rupture, thirteen of 16 had a live birth. 25 patients were treated with laparoscopic cornual pregnancy resection or cornual repair, and four out of 16 had a cornual rupture, twenty-two of 25 had a live birth. The live birth rate of cornual pregnancy reduction treatment was 81.2%, and the rate of cornual resection or repair was 85.6%. The live birth rate rates are similar for the two types of treatment.

One of the cases was a 34-year-old female patient with ultrasound and MRI suggesting a left cornual gestational sac measuring approximately 36*20*36mm and an intrauterine viable fetus at 13w+3days of gestational age [6]. Because of more severe pain, a diagnostic laparoscopy was performed. The left ovary and fallopian tube were not damaged and there were no signs of uterine rupture. The multidisciplinary team made a decision not to perform any further surgical interventions and keep monitoring. A live baby was delivered at 41 weeks of gestation.

Another patient with a diagnosis of cornual heterotopic pregnancy was confirmed on laparoscopy followed by methotrexate injection into the cornual gestational sac [7].

11 patients with cornual heterotopic pregnancy were treated with cornual pregnancy reduction under transvaginal ultrasound guidance, six of whom did not take feticide drug, four of them had a local injection of KCL(potassium chloride）, and 1 of them with a local injection of MTX（methotrexate） because the fetal heartbeat disappeared in utero [8]. One out of 10 patients treated with cornual pregnancy reduction, had uterine horn rupture and massive bleeding postoperatively, and ended up aborting. Eight out of 10 patients had a live birth. A retrospective cohort study enrolled 14 patients diagnosed with CHP. Six patients received cornual pregnancy reduction, and 2 had uterine horn rupture and massive bleeding which required emergency laparoscopic surgery for homostasis. No cornual rupture occurred among patients received laparoscopic cornual pregnancy resection. Each treatment group had one case of spontaneous miscarriage. The remaining 5 cases in cornual pregnancy reduction group and the remaining 7 cases in-aparoscopic cornual pregnancy resection group delivered healthy live offspring. In Wenzhi Xu ‘s study, 14 patients were treated with laparoscopic cornual pregnancy resection, and four out of 14 patients were confirmed to have ruptured corn, 3/4 as metrorrhagia [9]. One patient with ruptured spontaneous cornual heterotopic pregnancy at eight weeks of gestation underwent emergency laparotomy. The intrauterine gestational sac was accidentally ruptured because of its proximity to the ectopic pregnancy [10].

6. Discussion

Uterine horn pregnancies account for 1/7600 [11] of all pregnancies and 2%-3% of ectopic pregnancies. The horn pregnancy may abort spontaneously, may grow into the uterine cavity, though it may also grow outside of the uterine cavity, rupture and bleed from the thinning of the muscular layer of the horn tissue, with a maternal mortality rate of 2%-2.5% [12]. Differentiating between horn pregnancy and interstitial tubal pregnancy is a difficult problem for clinicians. Three-dimensional ultrasound or MRIs can more effectively diagnose these pregnancies [13]. The cornual pregnancy is located in the medial part of the round ligament, while the interstitial tubal pregnancy is located in the lateral part of the round ligament. Since most of the early stages are asymptomatic or involve only mild abdominal pain, heterotopic pregnancies are often detected by routine ultrasound. In Case 1, the local hospital ignored the patient’s embryo transfer of two embryos and ignored the uterine cornual gestational sac.

The cornual heterotopic pregnancy is an intrauterine pregnancy combined with a cornual pregnancy, which makes clinical management even more difficult in removing a horn pregnancy while aiming to ensure the safety of the intrauterine gestational sac. Cornual pregnancy reduction under transvaginal ultrasound guidance with or without the use of feticide drug and laparoscopic cornual pregnancy resection or cornual repair are the two most commonly used treatment modalities [14]. Expectant treatment is used only in a few cases such as when the fetal heartbeat of the gestational sac in the horn of the uterus disappears, the patient has no obvious abdominal pain, the gestational sac is not convex, etc.

Cornual pregnancy reduction under transvaginal ultrasound guidance is a relatively conservative form of treatment [3,9] though it is not indicated for patients with significant abdominal pain or a high risk of rupture. However, the risk of rupture is sometimes not accurately predicted. The trophoblast of a decompensated gestational sac is still active and the sac may still enlarge with time. Therefore, the size of the sac should be monitored by ultrasound regularly after surgery.

Cornual pregnancy resection or cornual repair is an effective treatment modality for CHP. The miscarriage rate of heterotopic pregnancy following treatment was approximately 6 to 33% [8]. In this study, the miscarriage rate of cornual pregnancy reduction treatment was 20%, and the miscarriage rate of cornual resection

or repair was 21.4%. Live birth rates are similar for the two types of treatment.

When performing a hysterectomy or repair procedures, two risks must have to be taken into account, The first of these risky wedge resection and complete extraction of the pregnancy increasing the risk of a blood loss during the procedure, and the potential risk of hysterectomy. The second risk is that the cornual resection may weaken the uterine musculature, increasing the risk of rupture during a subsequent pregnancy. This treatment may however, negate the complications of medically treated cornual pregnancy including the need for serial follow-up and the risks of delayed hemorrhage or rupture [7]. Patients with CHP who have severe pelvic adhesions are prone to horn rupture during laparoscopic separation of adhesions. Some CHP patients with a history of multiple previous surgeries or previous surgeries suggesting severe pelvic adhesions, or diseases such as pelvic tuberculosis and pelvic endometriosis, should be evaluated for the difficulty of surgery and the risks of rupture. Cornual pregnancy resection should be carefully planned particularly for the patients planning to have another child.

In Case 1, the patient had a history of two previous surgeries, namely one cesarean section and one bilateral salpingo-oophorectomy. During the second surgery, the tubes were enlarged and thickened bilaterally with severe pelvic adhesions. The treating physician opted for laparoscopic treatment because of the large gestational sac in the horn of the uterus. The patient bled 1500 ml during the operation and survived the intrauterine gestational sac. If the option of cornual pregnancy reduction under transvaginal ultrasound guidance was used with the feticide drug KCL, the patient may bleed significantly less. Laparoscopic monitoring during surgery, and prompt hysterectomy are required if there is significantly bleeding or the horn is ruptured. The purpose of laparoscopic monitoring is to stop the bleeding once intraoperative hemorrhage is detected; and the transvaginal puncture passes through less tissue and the operation is relatively simple. The uterus is less traumatized.

7. Conclusion

The management of CHP should take into account the type of cornual pregnancy, whether the cornual gestational sac is ectopic, whether the cornual gestational sac is ruptured and whether there are severe pelvic adhesions. For type I horn pregnancy, where rupture of the horn is unlikely, the transvaginal puncture is an option because it is less invasive. For the type II horn pregnancy, where the horn of the uterus is ectopic and the possibility of uterine rupture is high, laparoscopic surgery may be preferred. If the patient has had multiple previous surgeries, there may be dense adhesions in the pelvis and the corn of the uterus is prone to rupture during the separation of the adhesions, and laparoscopic surveillance with transvaginal sac puncture may be an option.

8. Conflicts of Interest Statement

The authors have no conflicts of interest relevant to this article.

9. Funding/Support Statement

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