

Surgical management of cornual heterotopic using intraoperative sonography depth resection guidance: case report

Allison Hagenow,¹ Sarah A Shaffer,² Surbhi C. Jain²

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Abstract

Heterotopic pregnancy is an uncommon phenomenon in which an ectopic and intrauterine pregnancy coexist. This condition can be life-threatening and poses a significant therapeutic challenge. Here we describe management of a heterotopic pregnancy with the ectopic pregnancy located in the right cornua. Resection of the ectopic pregnancy was performed via open laparotomy with intraoperative sonography. A sonography guided approach may optimize resection depth while secondarily allowing monitoring of the intrauterine pregnancy and prevention of disruption in cases in which the gestational sacs are in close proximity. After resection, the course of the pregnancy was uncomplicated, and a healthy baby was delivered via planned cesarean delivery at 36 weeks. While the optimal management of heterotopic pregnancies is often individualized, prompt diagnosis and treatment can result in favorable outcomes. The use of ultrasound intraoperatively allows for more precise resection depth, and which may lead to improved outcomes including increased intrauterine fetal survival rates and decreased

myometrial scarring.

¹*University of Iowa Carver College of Medicine, Iowa City, Iowa*

²*Department of Obstetrics and Gynecology, University of Iowa, Iowa City, Iowa*

Background

Ectopic pregnancy arises when an embryo implants outside of the intrauterine cavity and is a leading cause of maternal morbidity and mortality in the first trimester. The estimated incidence of ectopic pregnancy is 1-2% with the most common site of extrauterine implantation in the fallopian tube.^{1,2} Cornual ectopic is rare form of ectopic pregnancy occurring in an estimated 2-4% of all extrauterine pregnancies. Assisted reproductive technologies, such as in vitro fertilization and embryo transfer (IVF-ET), are established risk

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Corresponding authors: Allison Hagenow, Carver College of Medicine, University of Iowa, Iowa City, Iowa, allison-vaske@uiowa.edu. Sarah A. Shaffer, Department of Obstetrics and Gynecology, University of Iowa, Iowa City, IA, sarah-shaffer@uiowa.edu

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factors of ectopic implantation.¹⁻³

Heterotopic pregnancy (HP) occurs when an intrauterine pregnancy is present with at least one concomitant extrauterine development. In natural conception the incidence of HP is low occurring in one in every 30,000 pregnancies.^{3,4} This incidence is significantly increased with the use of IVF-ET with some theoretical models suggesting rates as high as one in every 119 pregnancies.^{3,4} This risk is further increased when multiple embryos are transferred during a single implantation procedure.

In this case report, we discuss a heterotopic cornual pregnancy after IVF transfer of two fresh embryos. Surgical resection of cornual ectopic via laparotomy resulted in continuation of a viable intrauterine pregnancy.

Case Report

The patient was a 37-year-old G2P1001 who presented to clinic for a consultation due to findings concerning for a cornual heterotopic pregnancy. Her first pregnancy was conceived via in vitro fertilization and resulted in a full term, uncomplicated vaginal delivery. At 11 months postpartum, she had two fresh embryos transferred in a single IVF cycle. Oocyte retrieval and fertilization occurred 5 days prior to transfer. At 6w3d a transvaginal ultrasound demonstrated a single viable intrauterine pregnancy. She then moved to a new city and at her first prenatal care appointment with a different provider, there was concern for a heterotopic pregnancy including the previously diagnosed intrauterine

pregnancy and a right cornual ectopic pregnancy. She was referred to our institution for further evaluation and presented to our clinic at 8w5d. Transvaginal ultrasound demonstrated Twin A appropriately positioned in the uterine cavity and Twin B visible to the right side of the uterus with concern for a cornual pregnancy. There appeared to be no myometrium between Twin B's gestational sac and the right adnexa, and the gestational sac did not appear to be communicating with the endometrial cavity. Given these findings, she was counseled on options of terminating both pregnancies versus an attempt at selective termination of the cornual pregnancy. She was counseled on the risks of the procedure including potential loss of both pregnancies. She accepted the risks and opted for selective termination. The next day, she was taken to the operating room where an exploratory laparotomy was performed and a cornual heterotopic pregnancy was confirmed [Figure 1].

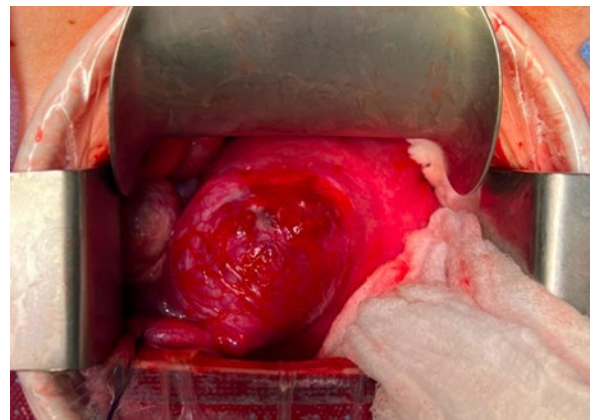


Figure 1: Ectopic pregnancy located in right cornua.

In addition to pre-operative transvaginal

ultrasound evaluation, intraoperative ultrasound was used to determine the

distance between the two gestational sacs (15 mm) [Figure 2].

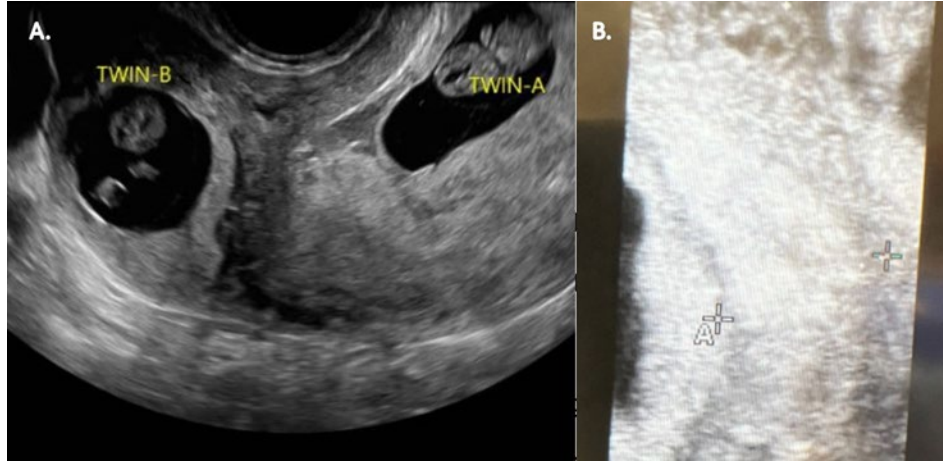


Figure 2: A. Pre-operative transvaginal sonographic image depicting gestational sacs of Twin A (intrauterine) and Twin B (ectopic). B. Intra-operative sonographic image depicting 15 mm between Twin A (intrauterine, left side of image) and Twin B (ectopic).

The cornual pregnancy was outlined with two purse-string stitches. Then cautery was used to incise the myometrium overlying cornual pregnancy. As the depth of dissection increased, gestational tissue began to spontaneously extrude from the cornua the purse string sutures were sequentially tightened. All gestational tissue was removed and sent to pathology. The serosa of the incision site was closed using a running, locked stitch of Chromic Gut suture [Figure 3].

Once hemostasis was achieved, ultrasound confirmed continued appropriate cardiac activity of the intrauterine pregnancy [Figure 4].

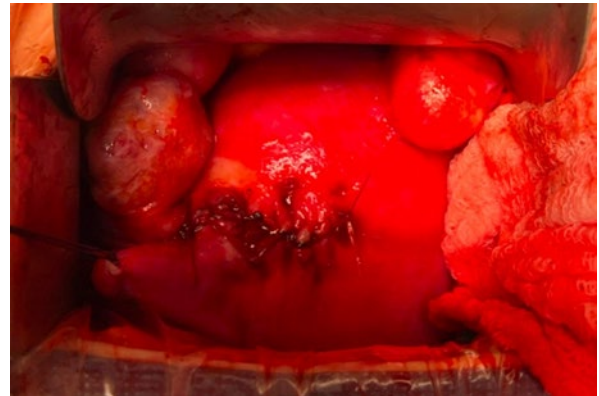


Figure 3: Final resection site after resection of gestational tissue and closure.



Figure 4: Intraoperative monitoring showing fetal cardiac activity in Twin A (intrauterine) status post resection of Twin B.

She subsequently had an uncomplicated prenatal course. It was recommended she deliver via cesarean section at 36 weeks which occurred without complication.

Discussion

The incidence of heterotopic pregnancy is overall rare but it increases with increased number of embryos transferred during a single IVF cycle.^{5,6} Decision for multiple embryo transfer is frequently influenced by maternal age as advanced maternal age is associated with lower pregnancy and live birth rates.⁵ Other factors that may influence number of embryos transferred include history of past IVF outcomes, financial considerations, embryo quality, and patient preference.^{5,7} To reduce the risks associated with multiple gestation pregnancies the American Society for Reproductive Medicine (ASRM, formerly The American Fertility Society) recommends limiting the number of embryos transferred during a single

cycle with an aim to promote transfer of a single gestation.⁷ The updated 2021 recommendations state that patients with a favorable prognosis should receive single embryo transfer if <35 years old, strong consideration should be made for single embryo transfer between 35-37 years old, and patients 38-40 years of age should have no more than 3 untested cleavage-stage embryos or 2 blastocysts transferred.⁷ Further recommendations are provided for patients with non-favorable scenarios. It is anticipated that these guidelines will continue to evolve as more information is obtained. The unique circumstances of each patient case along with a discussion of potential risk and preservation of patient autonomy should be principle in decisions regarding number of embryos transferred during an IVF cycle.

Heterotopic pregnancy poses a therapeutic challenge and, if untreated, can result in significant maternal morbidity and mortality in addition to the loss of one or both fetuses. Due to its rarity, the optimal management of heterotopic pregnancies remains controversial and should be individualized depending on clinical situation, patient stability, the size and site of the ectopic pregnancy, viability of the intrauterine pregnancy, physician expertise, patient preferences, available resources and other potential factors. Treatment options for heterotopic pregnancy include expectant management, medical therapies (i.e. methotrexate), ultrasound guided embryo aspiration (with or without injection of feticidal drugs), and surgical management with either laparotomy or laparoscopic approaches.^{1,8} One

retrospective study comparing surgical management, ultrasound-guided embryo aspiration, and expectant management in 52 patients with heterotopic pregnancy demonstrated worse maternal outcomes in expectant management including rupture of the ectopic pregnancy, need for medical facility transfer to facilitate surgical intervention due to continued growth of the heterotopic pregnancy and maternal fever.⁹

Medical treatments can be considered in stable patients with cornual ectopic pregnancies with initial low beta-hCG levels however, with a viable intrauterine pregnancy, medical management would be expected to cause termination of both developing embryos and would not be recommended if continuation of the intrauterine pregnancy were desired.^{1,6}

Ultrasound-guided transabdominal aspiration is considered a minimally-invasive option and has been demonstrated to have favorable maternal outcomes with low rates of loss of the intrauterine gestation. However, this is reserved for patients in which the gestational sac is unruptured and can be fully visualized pre-operatively.^{6,8,9} Laparoscopic surgical management has been increasing in popularity with the added advantage of complete removal of ectopic pregnancy.⁸⁻¹⁰ Laparotomy is classically reserved for ruptured ectopic pregnancy, the hemodynamically unstable patient, and/or significant hemoperitoneum.

Surgical management may confer a potential increased risk of early pregnancy loss of the intrauterine

pregnancy; however data has been inconsistent. One study demonstrated an increase in spontaneous abortion rates in surgical management compared to ultrasound-guided aspiration: 14.8% (4/27) compared to no abortions in the ultrasound aspiration group (0/5).⁹ These results should be considered with some scrutiny as the surgical management group was a larger cohort and the analysis included emergent surgical management which may not occur under ideal circumstances. Two of the four spontaneous abortions in the surgical cohort occurred in patients who presented with hemodynamically instability which may itself confer risk to continuation of the intrauterine pregnancy. Another retrospective study comparing outcomes in transvaginal ultrasound-guided aspiration versus laparoscopic resection of cornual heterotopic in 14 patients demonstrated similar spontaneous abortion rates between the two groups (16.7% vs 12.5%, p-value of 1.0).¹¹ Further data collection and analysis is needed to fully determine the differences in outcomes between these surgical approaches. Intraoperative ultrasound directly on the gravid uterus has similarly been described in a 2016 case report for cornual heterotopic resection.¹² The authors suggest that this approach allowed for improved guidance of suture planes for a more precise resection.¹² More investigation is necessary to determine the utility of intraoperative ultrasound guidance and its impact on maternal and fetal outcomes.

Conclusion

Our case describes an approach to surgical resection with the use of

intraoperative ultrasound to determine distance between intrauterine and cornual ectopic pregnancy. This approach improved accuracy of required depth of resection. One consequence of surgical cornual resection is development of a uterine scar which can become a site of uterine rupture during labor or future pregnancies.¹³ Intraoperative ultrasound could serve as a valuable intraoperative tool to reduce the size and depth of resection therefore minimizing the extent of uterine scarring and improving maternal and fetal outcomes following cornual ectopic pregnancy resection in the setting of a heterotopic gestation.

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