Heterotopic cervical pregnancy successfully treated with transvaginal ultrasonic-guided aspiration and cervical-stay sutures

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**Objective:** To present a case of a heterotopic cervical pregnancy successfully treated with transvaginal ultrasonic-guided aspiration and cervical-stay sutures.

**Design:** Case report.

**Setting:** Tertiary academic IVF program.

**Patient(s):** A 35-year-old woman who conceived from IVF-ET treatment at 5.5 weeks of gestation.

**Intervention(s):** Transvaginal ultrasound–guided aspiration of the cervical pregnancy followed by cervical-stay sutures to control hemorrhage after aspiration.

**Main Outcome Measure(s):** Recovery of the patient, preservation of the intrauterine pregnancy, and sequelae.

**Result(s):** The cervical pregnancy was successfully aborted, and the intrauterine pregnancy progressed to term.

**Conclusion(s):** Transvaginal ultrasound–guided aspiration in combination with hemostatic cervical-stay sutures can be safely used to manage heterotopic cervical pregnancies. (Fertil Steril 2001;75:1030 –3. ©2001 by American Society for Reproductive Medicine.)

**Key Words:** Heterotopic pregnancy, cervical pregnancy, transvaginal ultrasound-guided aspiration, cervical-stay sutures

Cervical pregnancy is a rare, potentially life-threatening condition. The incidence has been reported to be between 1 of 1000 and 1 of 18,000 pregnancies (1). Although the etiology of cervical pregnancy is unclear, predisposing risk factors have been identified, including prior operative uterine procedures (i.e., dilatation and curettage, cesarean section), use of an intrauterine device, and in vitro fertilization (2). Because of the vascularity and lack of muscular tissue in the cervical region, catastrophic hemorrhage often occurs in undiagnosed and untreated cervical pregnancies. Hemorrhage frequently requires life-saving hysterectomy, at the cost of any future reproductive potential.

In 1978, Raskin reported the first sonographic diagnosis of a cervical pregnancy (3). Since then, the mainstream use of ultrasound has enabled early diagnosis, and several conservative management options have since been observed. These options range from curettage and packing to methotrexate, angiographic embolization, and ultrasound-guided fetocidal injections (4).

Heterotopic cervical pregnancy is an even rarer condition, and treatment is frequently complicated by the desire to maintain the intrauterine pregnancy. Conservative management techniques in these cases generally focus on selective fetal reduction of the cervical pregnancy using ultrasound-guided intra-amniotic/cardiac injections of potassium chloride (KCI; see References (5–7)). We present a case of an early heterotopic cervical pregnancy in which the cervical pregnancy was terminated using transvaginal ultrasound-guided aspiration. The cervical pregnancy aborted 16 days
later, and cervical-stay sutures were used to achieve hemostasis. The intrauterine pregnancy progressed to term without further complication. To our knowledge, this is the first reported case of the successful use of transvaginal ultrasound-guided pregnancy aspiration and hemostatic cervical-stay sutures in a heterotopic cervical pregnancy.

**CASE REPORT**

A 35-year-old woman, Gravida 1, Para 0, (G1P0), with a diagnosis of tubal-factor infertility and asthenoteratozoospermia in the male partner who had been unable to conceive for 1.5 years underwent IVF at our institution. The patient’s history was notable for a laparoscopy/hysteroscopy 3 months before IVF, at which time a bilateral salpingectomy was performed for bilateral hydrosalpinges. An endometrial polyp was also removed at that time.

Her IVF stimulation protocol included low-dose luteal leuprolide acetate down-regulation, followed by a daily administration of 2 ampules of recombinant hFSH plus 2 ampules of hMG. The patient’s E₂ level reached a peak of 1,823 pg/mL on cycle day 14, at which time 5,000 units of hCG was administered. Four of five retrieved oocytes were successfully fertilized with ICSI. Three days after oocyte retrieval, all four embryos (9-, 8-, 8-, and 7-cell embryos, respectively) were transferred without difficulty using a Wallace catheter. Luteal-phase support in the form of 50 mg/d intramuscular progesterone was given. The initial serum hCG level was 230.3 mIU/mL 11 days after embryo transfer (ET) and was then followed weekly over the next 2 weeks (5,154 mIU/mL 18 days after ET; 28,910 mIU/mL 25 days after ET).

At the time of her last hCG level, 25 days after ET, the patient reported some mild vaginal bleeding. Transvaginal ultrasound was performed, revealing a live twin gestation, with one sac measuring 10 × 6 mm [fetal crown rump length (CRL) = 4.4 mm] located in the cervical canal just below the level of the internal os, with positive fetal heart movement. Doppler vascular blood flow to this area confirmed the suspected heterotopic cervical pregnancy. Pelvic examination revealed a closed cervix and a small amount of blood in the vaginal vault without any signs of active bleeding.

The patient was then admitted to the hospital. Transvaginal ultrasound-guided aspiration of the cervical pregnancy was performed using a 16-gauge Cook needle. Three milliliters of KCl was then injected into the region of the disrupted cervical pregnancy. The patient tolerated the procedure well without any excessive bleeding. During the procedure, interventional radiologists were on standby to perform uterine artery embolization if necessary. The patient was observed overnight and discharged the following day.

Histologic inspection of the aspirated tissue showed chorionic villi. Ultrasound performed 3 days after the procedure revealed a 5-mm hypoechoic area in the cervix and confirmed the viability of the intrauterine pregnancy. Sixteen days after the procedure, the patient experienced vaginal bleeding. Further examination revealed aborting tissue and significant active bleeding. Under epidural anesthesia, hemostatic synthetic absorbable sutures were placed high on the cervix at 1, 3, 9, and 10 o’clock, ultimately circumferentially tying the cervix (Fig. 1A). The aborting tissue that was bulging through the cervical os was then removed, and the sutures at 3 and 9 o’clock were tied together, thus achieving complete hemostasis (Fig. 1B).

The intrauterine pregnancy proceeded without further complication. The cervical-stay sutures dissolved by the 18th–20th weeks of gestation. There was no evidence of cervical incompetence throughout the remainder of the pregnancy. At 38 weeks of gestation, the patient delivered a liveborn male infant weighing 3,345 g via cesarean section for cephalopelvic disproportion. At the time of the surgery, the cervical canal was carefully inspected and noted to be normal.

**DISCUSSION**

The incidence of heterotopic pregnancies has increased as the use of in vitro fertilization has become more widespread. Heterotopic cervical pregnancies, although still extremely rare, would be expected to increase in frequency with the expanding use of assisted reproductive technologies (ART). The main cause of the increased incidence of cervical pregnancy in ART is unclear. It may be a result of infertility factors or secondary to the IVF procedure itself. Cervical trauma or embryo reflux into the cervix during embryo transfer are two popular hypotheses (1).

The diagnosis of cervical pregnancy has been classically established through pathologic and clinical means. The presence of villi in endocervical stroma would constitute pathologic proof (8). Clinically suggestive criteria include an hourglass-shaped uterus; a softened, disproportionately enlarged cervix in relation to the uterus; and/or products of conception entirely confined within and firmly attached to the endocervix (8). Kligman et al. (9) reviewed the diagnostic dilemma that frequently occurs in treating cervical pregnancies. Unfortunately, these diagnostic criteria are usually made at a greater gestational age, in emergent situations when it is too late for conservative measures.

Parente et al. (10) estimated that cervical pregnancies that have progressed beyond 10 weeks of gestation tend to bleed so profusely (either spontaneously or on evacuation) as to require hysterectomy for control. The advent of transvaginal sonography and magnetic resonance imaging has made the early diagnosis of cervical pregnancy possible. Ultrasound, in particular, is now the method of choice for diagnosing cervical pregnancy. Placental implantation in the cervix, internal echoes within an enlarged cervical canal, and an hourglass-shaped uterus are all ultrasound findings sugges-
tive of cervical pregnancy. In this case, the diagnosis was further confirmed through the use of Doppler sonography, which revealed significant blood flow to the suspected cervical pregnancy. Clearly, a cervical abortion (an abortion in which the products of conception are retained in the cervical canal by a resistant external os) would not have demonstrated a vascular relationship with the cervix.

Although there has been considerable debate as to the true incidence of cervical pregnancies given some of the uncertainties in diagnosis, the treatment of cervical pregnancy has progressed. Maternal mortality has decreased from 40% in 1911 to <10% by 1953 to almost 0 today (10, 11). Up until 1979, an estimated 89% of cervical pregnancies required emergent abdominal hysterectomies (12). Conservation of the uterus was achieved by combining curettage with cervical packing, suturing, balloon tamponade, and uterine artery ligation/embolization. Conservative management by cervical cerclage was first described by Scott et al. in 1978 (13). Bernstein et al. (14) describes two further cases where hemostasis was achieved with a McDonald type cervical suture and vaginal packing. Medical management with methotrexate has been shown to be a viable option (1). Successes with methotrexate have been reported as late as 13 weeks into gestation.

There have been several case reports of heterotopic cervical pregnancies in the literature. Bratta et al. (15) reported successfully using intrauterine methotrexate to treat a heterotopic cervical pregnancy. Ginsburg et al. (2) described a triplet heterotopic gestation with a twin cervical pregnancy, which was treated with uterine artery embolization and dilatation and evacuation. In both cases, the intrauterine pregnancy was also aborted. Davies et al. (16) used digital evacuation of a cervical pregnancy and cervical cerclage at 10 weeks into gestation in an attempt to control cervical hemorrhage while maintaining the intrauterine pregnancy. However, demise of the intrauterine pregnancy occurred at 13 weeks of gestation.

Monteagudo et al. (5) and Frates et al. (6) describe ultrasound-guided injection of KCl into the cervical pregnancy in the first trimester, followed by cesarean section after 34 weeks for the intrauterine gestation. In both cases, no attempt was made to aspirate the cervical pregnancy. Although there was no longer any fetal cardiac activity in the cervical pregnancy, the cervix remained hypervascular throughout the pregnancy, necessitating cesarean section to deliver the intrauterine fetus in each case. Subsequently, methotrexate injections were used to treat the remaining cervical pregnancy in the postpartum period.
Honey et al. (7) performed fluoroscopic uterine artery embolization in addition to ultrasound-guided intracardiac KCl to control bleeding in a heterotopic cervical pregnancy. The intrauterine pregnancy survived the initial treatments; however, chorioamnionitis developed approximately 1 week after the patient was discharged, and the patient eventually underwent a total abdominal hysterectomy.

This is the first reported case of the use of transvaginal ultrasound-guided aspiration and cervical-stay sutures to abort a cervical pregnancy in the setting of a concomitant viable intrauterine pregnancy. Clearly, several factors played a role in choosing this course of management. Profuse bleeding would have precluded this type of treatment. The fact that this pregnancy was conceived via IVF increased the vigilance and early monitoring. The small size and early diagnosis of the cervical pregnancy, as well as operator proficiency with ultrasound-guided aspiration, were major considerations.

In contrast to the other case reports regarding cervical heterotopic pregnancies, aspiration of the cervical pregnancy was performed in this instance. Aspiration of the cervical pregnancy carries the risk of hemorrhage. This risk increases greatly with increasing gestational age, and as this case demonstrated, cervical-stay sutures were necessary to control hemorrhage that occurred 16 days after the aspiration. This suggests that perhaps the aspiration and intracervical KCl injection were sufficient to disrupt the cervical pregnancy but were not enough to completely evacuate it. Other reports have described how the remaining cervical pregnancy tissue has continued to implant and develop or act as a nidus for infection.

The attractiveness of conservative management of heterotopic cervical pregnancies is understandably high, especially in cases involving ART. Therefore, risk tolerance levels increase, but undoubtedly, maternal safety must always be paramount. A procedure that completely evacuates the cervical pregnancy without undue risk of trauma to the intrauterine pregnancy, hemorrhage, or infection would be ideal. This case illustrates that in the appropriate setting, aspiration of a cervical pregnancy, followed by cervical-stay sutures when necessary, can be safely undertaken.

References