

# Management of a cervical heterotopic pregnancy presenting with first-trimester bleeding: case report and review of the literature

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**Objective:** To report a rare case of a cervical heterotopic pregnancy resulting from intrauterine insemination (IUI) that presented with first-trimester bleeding.

**Design:** Case report and literature review.

**Setting:** Large university-affiliated infertility practice.

**Patient(s):** A 40-year-old gravida 2 para 1 Asian woman at 7-3/7 weeks gestational age following clomiphene citrate/IUI for the treatment of secondary infertility presented with heavy vaginal bleeding for several days.

**Intervention(s):** Transvaginal ultrasound on admission revealed a single live intrauterine pregnancy and a cervical gestational sac containing a nonviable embryo. The patient continued to have vaginal bleeding and 2 days later underwent removal of the cervical ectopic pregnancy tissue with ring forceps, as well as an ultrasound-guided intracervical Foley balloon and cerclage placement. The bleeding subsided, and 48 hours later the Foley and cerclage were removed.

**Main Outcome Measure(s):** Pregnancy outcome.

**Result(s):** The remainder of the pregnancy was uncomplicated and the patient had a full-term cesarean delivery for footling breech of a healthy male infant.

**Conclusion(s):** Cervical heterotopic pregnancy is a very rare event that almost universally results from infertility treatment. We present a case where we were able to remove the cervical ectopic and tamponade the bleeding, thus preserving the intrauterine pregnancy for this subfertile couple, and we review the existing literature. (Fertil Steril® 2012;98:89-94. ©2012 by American Society for Reproductive Medicine.)

**Key Words:** Cervical pregnancy, heterotopic pregnancy, Foley balloon, cervical cerclage

The widespread use of assisted reproductive technologies (ART) has increased the frequency of certain rare and potentially life-threatening pregnancies with abnormal implantation (1). The incidence of heterotopic pregnancies is as high as 1% in some studies (2) and cervical ectopic

pregnancies occur in 1 in 2,500-18,000 pregnancies, mostly as the result of ART (3).

Cervical heterotopic pregnancy is an even less frequent condition that refers to the coexistence of at least one gestational sac in the uterus and at least one in the cervical canal. The diagnosis

is mainly based on sonographic findings. There are currently no standard treatment guidelines, and each case is approached individually based on patient preference, physician experience, and resource availability.

We report the case of a cervical heterotopic pregnancy, in which the intrauterine component was a single viable gestation. The patient presented with first-trimester bleeding and was managed successfully. Previously published cases are also reviewed.

## CASE REPORT

A healthy 40-year-old gravida 2 para 1 Asian woman presented to our infertility center for evaluation of secondary

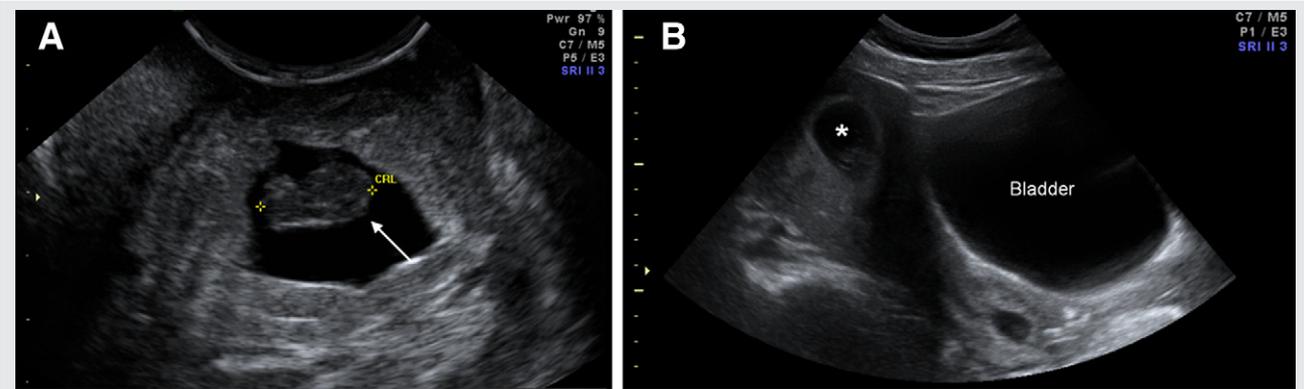
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FIGURE 1



Transabdominal ultrasound images demonstrating nonviable cervical pregnancy (arrow) (A) alone or (B) along with intrauterine pregnancy (asterisk). CRL = crown-rump length.

Moragianni. Cervical heterotopic pregnancy. *Fertil Steril* 2012.

infertility. Her past obstetrical history was significant for an uncomplicated term vaginal delivery, followed by a 6-week spontaneous miscarriage that was treated with dilatation and evacuation. The patient had been attempting conception with the same partner for the past 3 years. Her past medical and surgical history was otherwise unremarkable and the infertility evaluation of both her and her male partner did not reveal any abnormalities. The patient subsequently underwent two cycles of ovulation induction with clomiphene citrate and intrauterine insemination (IUI), the first of which did not result in pregnancy. After the second IUI cycle, the patient presented with several episodes of heavy vaginal bleeding, and initial ultrasounds revealed a single live intrauterine pregnancy. At 7-3/7 weeks' gestation a pelvic ultrasound revealed a single live intrauterine pregnancy as well as a cervical ectopic pregnancy without fetal cardiac activity (Fig. 1). The patient was subsequently admitted to the hospital owing to the persistence of vaginal bleeding and the need for management of her heterotopic gestation. She was extensively counseled regarding her options of termination versus conservative management and opted for what was deemed the most intrauterine pregnancy-conserving approach. On the first hospital day, at 7-5/7 weeks' gestation, she was taken to the operating room and the interventional radiology service was alerted and available in case the need for embolization arose. The cervical ectopic was removed under ultrasound guidance with ring forceps. The patient then developed heavy vaginal bleeding, and placement of a vaginal bivalve speculum allowed visualization of the bleeding tissue arising from the endocervix, with extension slightly beyond the cervical os. Application of gentle pressure to the tissue with ring forceps revealed it to be closely adherent to the wall of the endocervical canal. Tamponade was achieved with ultrasound-guided placement of an endocervical Foley catheter (5 mL Foley balloon placed to a depth of 2 cm), which was secured in place with double mattress cerclage sutures of the anterior cervix (Fig. 2). Use of intraoperative transvaginal and transabdominal sonography performed by a skilled ultrasonologist allowed precise measurements to accurately guide

the placement of the catheter balloon's maximal pressure point against the bleeding vessels. This technique was enhanced with Doppler flow assessments done before and after achieving successful tamponade of the endocervical placental bed and prevented mechanical interruption of the intrauterine placental tissue. At the completion of the procedure, abdominal and transvaginal ultrasound confirmed intrauterine fetal cardiac activity, as well as minimal Doppler flow and pooling of blood at the cervical ectopic bed. The estimated blood loss for the procedure was 150 cc and the patient was not given antibiotics. Two days later, no further bleeding was noted and we removed both the cerclage suture and Foley catheter without difficulty. The patient was discharged home and was advised to avoid intercourse during the first trimester of the pregnancy. Pathologic examination of the tissue removed from the cervix confirmed the presence of chorionic villi. The patient was monitored closely for the remainder of

FIGURE 2



Transabdominal ultrasound images demonstrating Foley catheter balloon (star) and cerclage (arrow) placed in cervix.

Moragianni. Cervical heterotopic pregnancy. *Fertil Steril* 2012.

TABLE 1

## Overview of published cervical heterotopic cases (presented in reverse chronologic order of publication).

	Ref.	GA at diagnosis	Mode of conception	Treatment	Outcome
1	Current report	7w3d	CC/IUI	Extraction of cervical pregnancy, placement of Foley (5 ml) and cerclage (48 hrs)	Delivery (FT, c/s)
2	(4)	6w5d	IVF	Bilateral uterine artery MTX injection and embolization with gelatin sponge	Termination
3	(5)		Spontaneous	Single-dose intra-amniotic MTX injection in the gestational sac	Delivery (FT, vaginal)
4	(6)	5w5d (1 nonviable intrauterine, 2 cervical)	IVF/DO	D&C of all 3 pregnancies	Termination
5	(1)	9w	CC	Cervical D&C	Delivery (FT)
6	(7)	6w	IVF	Foley placement for 3 days, cerclage, ligation of descending cervical branches of the uterine arteries, MTX injection, D&E of intrauterine pregnancy	Termination
7	(8)	6w	IVF	KCl injection of cervical pregnancy	Delivery (31w, c/s)
8	(9)	6w	IVF	Cervical D&C (ultrasound-guided)	Delivery (FT, c/s)
9	(10)	8w	Spontaneous	Aspiration of cervical pregnancy, placement of Foley (6-Fr, 3 mL, 24 h)	Delivery (FT, c/s)
10	(11)	7w	IVF/ICSI	Aspiration of cervical pregnancy (ultrasound-guided), bilateral hypogastric artery balloon placement	Delivery (FT, c/s)
11	(12)	7w3d	IVF	Aspiration of cervical pregnancy (ultrasound-guided)	Delivery (35w, vaginal)
12	(13)	6w (2 intrauterine, 1 cervical)	IVF	Aspiration and KCl injection of cervical pregnancy (ultrasound-guided), ligation of descending cervical branches of uterine arteries	Doing well at 12w
13	(14)	6w (2 intrauterine, 1 cervical)	IVF	Aspiration and hyperosmolar glucose injection of cervical pregnancy (ultrasound-guided)	Delivery (34w, c/s), postpartum hemorrhage, large cervical hematoma
14	(15)	7w (1 intrauterine, 2 cervical)	IVF	Selective intrauterine artery catheterization, MTX injection and embolization with Gelfoam	Termination
15	(16)	6w (2 intrauterine, 1 cervical)	IVF	Aspiration of cervical pregnancy	Delivery, twins (29w, c/s)
16	(17)	6w	IVF	KCl injection of cervical pregnancy (ultrasound-guided)	Delivery (31w), cesarean hysterectomy
17	(18)		IVF	Kleppinger bipolar electrocautery on external os without cervical dilatation (ultrasound-guided)	Delivery (FT, vaginal)
18	(19)	7w	Spontaneous	KCl injection of cervical pregnancy (ultrasound-guided)	Delivery (35w, c/s), cervical canal hemorrhage managed with sutures
19	(20)	8w	IVF	Extraction of cervical pregnancy and gauze compression	Delivery (FT, vaginal)
20	(21)	5w (2 intrauterine, 1 cervical)	IVF	Aspiration and hyperosmolar glucose injection of cervical pregnancy	Delivery, twins (34w, c/s)
21	(22)	6w	Spontaneous	Aspiration of cervical pregnancy (ultrasound-guided)	Miscarriage
22	(23)	6w	IVF/ICSI	Intramuscular cervical layer Pitressin injection, transvaginal extraction of cervical pregnancy with forceps and curettage	Delivery (FT, VAVD)
23	(24)	12w	IVF	Aspiration and KCl injection of cervical pregnancy	Delivery (36w), cesarean hysterectomy, DIC

Moragianni. Cervical heterotopic pregnancy. Fertil Steril 2012.

TABLE 1

Continued.

	Ref.	GA at diagnosis	Mode of conception	Treatment	Outcome
24	(25)	7w	IVF/ICSI	Hysteroscopic and rollerball electrocautery of cervical pregnancy, McDonald cerclage (12w)	Delivery (FT, c/s)
25	(26)	5w (2 intrauterine, 1 cervical)	IVF	Transvaginal extraction of cervical pregnancy with forceps (ultrasound-guided)	Delivery (FT, c/s)
26	(27)	8w3d	IVF	Shirodkar cerclage	Delivery (FT)
27	(28)	6w	IVF	Aspiration and KCl injection of cervical pregnancy (ultrasound-guided), cerclage	Delivery (FT, c/s)
28	(29)	5w	IVF	Spontaneous resolution of cervical pregnancy at 5+w	Delivery (FT)
29	(30)	6w	IVF	KCl injection of cervical pregnancy	Delivery (36w)
30	(31)	6w	IVF	KCl and MTX injection of cervical pregnancy (ultrasound-guided)	Delivery (30w, c/s)
31	(32)	8w	IVF	Uterine artery embolization, KCl injection of cervical pregnancy	Miscarriage, hysterectomy (vaginal bleeding and infection)
32	(33)	9w6d	FSH/UII	KCl injection of cervical pregnancy	Delivery (34w, c/s)
33	(34)	7w		MTX injection × 2	Termination
34	(35)		IVF	KCl injection of cervical pregnancy	Delivery
35	(36)	7w	IVF		Miscarriage
36	(37)	7w	OI	KCl injection of cervical pregnancy	Delivery (35w)
37	(38)	7w	IVF	Selective intrauterine artery catheterization and MTX injection	
38	(39)	10w	IVF	Digital evacuation and cervical cerclage	Miscarriage (13w)
39	(40)	8w	IVF	D&E of both pregnancies	Termination

Note: CC = clomiphene citrate; c/s = cesarean section; D&C = dilatation and curettage; D&E = dilatation and evacuation; DO = donor oocyte; FT = full term; GA = gestational age; ICSI = intra-cytoplasmic sperm injection; IUI = intrauterine insemination; MTX = methotrexate; OI = ovulation induction; VAVD = vacuum-assisted vaginal delivery.

Moragianni. Cervical heterotopic pregnancy. *Fertil Steril* 2012.

the pregnancy, which progressed uneventfully and resulted in the birth of a healthy male infant at 39 weeks by cesarean section for footling breech presentation.

## DISCUSSION

Cervical ectopic pregnancies account for <1% of all ectopic pregnancies (1). Cervical heterotopic pregnancies are more infrequent, despite a slightly increased incidence as a result of recent ART practices. Risk factors include uterine and cervical anomalies, prior uterine curettage or cesarean delivery, smoking, tubal-factor infertility, and IVF treatment (4). Patients typically present with vaginal bleeding, with or without abdominal/pelvic pain. In addition to a thorough physical examination, pelvic (preferably transvaginal) sonography is used to confirm the diagnosis.

We performed a Medline search using the key words “heterotopic pregnancy” and “cervical” and identified 57 records, of which there were 38 reported cases of cervical heterotopic pregnancy besides the one presented here (Table 1). Currently, there are no guidelines for the treatment of cervical heterotopic pregnancies. It is therefore imperative to provide extensive counseling for the patient, because there is no evidence to support the use of any one therapeutic modality. Of these 39 cases, four were the result of natural conception and five resulted from ovulation induction. As expected, the majority of

cases (n = 30; 77%) were conceived with IVF. The gestational age at diagnosis ranged from 5 to 10 weeks, and all cases were diagnosed based on sonographic findings. Regarding treatment modalities, surgical removal of the cervical pregnancy (aspiration, extraction, dilatation and curettage, hysteroscopy) and medical management (injection of methotrexate, potassium chloride, or hyperosmolar glucose) were used in the majority of cases (n = 16 [41%] and n = 14 [36%], respectively), with a smaller proportion of cases utilizing a combination of treatment modalities (n = 7; 18%). Three of the reported cases (8%) describe the use of a Foley catheter, and six (13%) of a cervical cerclage. In two of these cases (5%) the authors describe using both a Foley catheter and a cervical cerclage. Embolization or ligation of the uterine arteries by various modalities was reported in seven cases (18%). In only one of the 39 cases (3%) was spontaneous resolution of the cervical ectopic pregnancy reported.

Because the majority of these pregnancies result from use of ART in the subfertility population, patients are even more likely to desire preservation of both the intrauterine pregnancy and their future fertility. Of the 39 reported cases, 27 (69%) resulted in delivery of the intrauterine gestation, two of which were sets of liveborn twins. Of all reported deliveries, 14 were full term (52%), and 12 were preterm (44%), with preterm delivery gestational ages ranging from 29 to 36 weeks. Of those pregnancies with a reported method of delivery (n

= 21), the majority were cesarean (n = 16; 76%) versus vaginal (n = 5; 24%). Several of these cases had fetal indications for cesarean delivery, but this reason alone would not necessarily account for such a high cesarean section rate. It is possible that a higher incidence of placental abnormalities or residual trophoblastic tissue causing bleeding might also account for this finding. Six of the cases were electively terminated (15%), whereas four resulted in spontaneous abortion (10%). Three of the 39 cases (8%) were complicated by hysterectomy owing to excessive bleeding, a devastating outcome for any reproductive-age woman.

This literature review was limited by the small number of published case reports, preventing a consensus on definitive success rates or treatment recommendations. Observations were further limited by reporting bias, because negative treatment outcomes are less likely to be submitted or accepted for publication. In addition, the paucity of reported cases does not allow a determination of superior management approach, i.e., surgical versus medical. In those cases in which access to the ectopic pregnancy is impaired, management with methotrexate, when applied locally or intra-amniotically, may be preferable. However, because of the viable intrauterine pregnancy, monitoring via ultrasound (as opposed to hCG levels) is required to ensure treatment success.

In the case we report here, the cervical pregnancy was removed with the use of ring forceps, and mechanical tamponade of the cervical bed, with the use of a Foley catheter and cervical cerclage, was maintained for 48 hours until hemostasis was assured. The intrauterine gestation, as well as the patient's potential future fertility were preserved. This particular case could be best managed surgically owing to easy access to the cervical bed and precise sonographic guidance. It is imperative that accurate ultrasound images are obtained, necessitating multiple ultrasounds and the need for repeated confirmatory testing, especially in high-risk patients.

In conclusion, when the clinician is faced with a cervical heterotopic pregnancy, the patient's status and wishes should be assessed, as well as the physician experience and availability of resources. In the 39 reported cases reviewed, fertility-sparing management appears to be an option that will likely result in the viable delivery of the intrauterine gestation, without compromising the patient's reproductive capacity.

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