



Uterine Artery Embolization in the Twentieth Week of Pregnancy in Abnormally Invasive Placenta with Live Birth

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ABSTRACT

An abnormally invasive placenta is an increasing and potentially life-threatening pregnancy complication. The case presented herein is a heterotopic dichorial pregnancy with implantation of 1 placenta within the isthmocervical area, which caused vaginal bleeding during the 20th week of pregnancy, requiring a blood transfusion. To stop the bleeding, a bilateral embolization of the cervical branches of the uterine arteries was performed. The embolization was well tolerated and resulted in the abrupt and lasting cessation of bleeding for more than 10 weeks, resulting in the live birth of 1 child.

ABBREVIATIONS

AIP = Abnormally invasive placenta, uterine artery embolization = (UAE)

Abnormally invasive placenta (AIP) is an increasing and potentially life-threatening pregnancy complication (1). Due to a defect within the decidua (endometrium), the trophoblast (placental cells invading the uterus to enable implantation) expands into the myometrium and can infiltrate adjacent pelvic organs. In the presented case, a cervicoisthmic implantation occurred, defined as implantation of the gestational sac partially within the endocervix and partially within the caudal uterine decidua (2). Risk factors for complete or partial cervical implantation are previous uterine procedures (3, 4) and curettages (2), which can cause decidual damage and pave the way for AIP development (5). In these cases, severe

bleeding occurs when the placenta is detached, often resulting in hysterectomy as the only effective treatment option (6). Live births in patients with isthmocervical implantation have been reported previously; however, severe bleeding after placental detachment due to AIP provoked intrapartum-hysterectomy in many cases (7, 8).

The present study reports a case of 1 pregnant patient with 2 fetuses and a heterotopic dichorial (twins with 1 amniotic sac and placenta each) pregnancy within the isthmocervical area. Recurring and steadily increasing vaginal bleeding required blood transfusion during the 20th week of pregnancy. Embolization of cervical branches of both uterine arteries (9, 10) resulted in abrupt and maintained cessation of bleeding over 10 weeks resulting in the live birth of 1 fetus.

MATERIALS AND METHODS

Exemption from ethics approval and waiver of informed consent were granted by the ethics committee.

Patient

A 39-year-old patient with 5 gravidae (pregnancies, completed and incomplete) and 2 paras (completed pregnancies) was previously treated with a total of 5 curettages due to residual placental tissue postpartum and first trimester miscarriages. The patient desired to have 1 more child.

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None of the authors have identified a conflict of interest.

IRB approval status: Exemption from ethics approval and waiver of informed consent were granted by the ethics committee.

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J Vasc Interv Radiol 2021; 32:339–342

<https://doi.org/10.1016/j.jvir.2020.12.002>

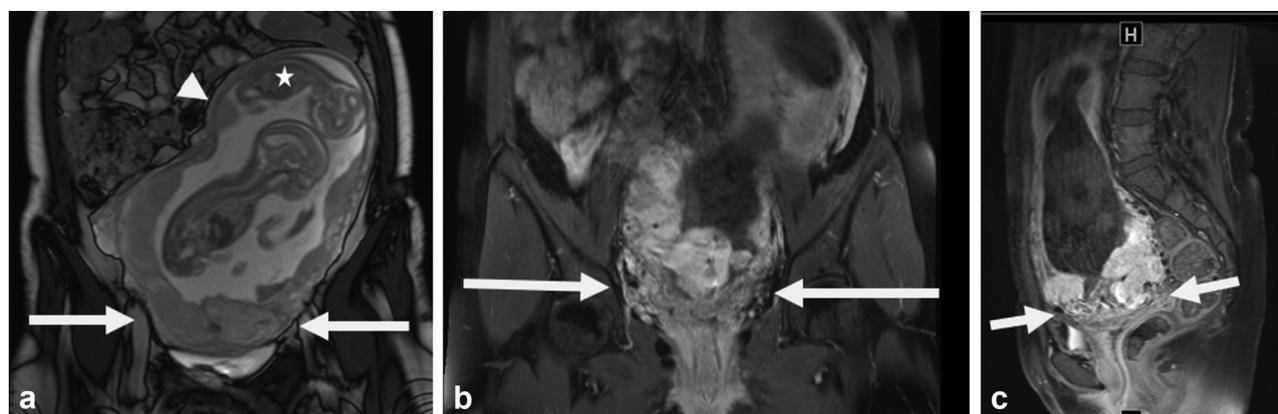


Figure 1. MR image of a 39-year-old woman during the 20th week of pregnancy. (a) Coronal T2w image showing the placenta extending from cranial (arrowhead) to isthmicocervical (arrows) and 2 fetuses with different gestational ages, 1 with appropriate gestational age, and 1 with intrauterine growth retardation (star). Contrast-enhanced T1w images with fat-suppression in (b) coronal and (c) sagittal orientation visualize the hyperperfused region at the isthmicocervical junction (arrows).

Clinical and laboratory investigation revealed that the anti-Müllerian hormone was low (0.22 ng/mL; normal level of fertile women, 1.0–4.0 ng/mL), and oligomenorrhea resulted from premature ovarian failure. Additionally, the endometrium was thin (<5 mm), even at the time of ovulation. Although an Asherman syndrome (adhesions inside the uterus) could be excluded by hysteroscopy, endometrial damage was suspected. Thus, ovarian stimulation treatment with menotropin was started and successfully resulted in dichorial twin pregnancy.

After 14 weeks and 1 day of pregnancy (technical term = 14+1 weeks, used hereafter), the patient presented with vaginal bleeding. Clinical examination revealed an open cervix with visible placental tissue. The sonographic examination further showed implantation of 1 placenta within the cervicoisthmic region, whereas the other was implanted within the cavum uteri. The caudal placenta within the cervicoisthmic region infiltrated nearly the complete endocervix. Ultrasound also showed the full spectrum of AIP: lacunae, extensive vascularization, nonvisible myometrial wall.

While under inpatient surveillance, a cervical cerclage was performed to avoid progressive bleeding due to the dilating cervix. After 16+5 weeks of pregnancy, sonographic examination showed growth differences between the fetuses with intrauterine growth retardation of the intracavum fetus. Moreover, the patient experienced increasing vaginal bleeding. After 19+3 weeks of pregnancy, she presented with a hemoglobin level of 7.7 g/dL, and a blood transfusion was performed. As the patient refused to consent to a hysterectomy, embolization of the uterine arteries as an ultima ratio treatment option was considered. The patient was informed about missing clinical data and the unforeseeable consequences for the viable fetus. On the other side, the intracavum fetus showed progressive signs of severe placental insufficiency. Pre-eclampsia marker soluble fms-like tyrosine kinase-1/placental growth factor increased substantially to a maximum ratio of 151 (>85 represents a pathologic ratio in

the gestation phase until week 34). The patient consented to uterine artery embolization (UAE).

Uterine Artery Embolization

A preprocedural magnetic resonance (MR) imaging was performed to visualize the anatomical situation in relation to the cervix and the uterine artery. Moreover, imaging depicted the position of both fetuses (Fig 1a). Contrast-enhanced imaging demonstrated isthmic hyperperfusion of the placenta, the target for selective embolization (Fig 1b, c). UAE was performed at 19+4 weeks of pregnancy with local anesthesia. During angiography, the uterine artery was catheterized sequentially on both sides using a 4-F catheter (Glidecath; Terumo, Somerset, New Jersey) in the vertebral configuration. A coaxially introduced microcatheter (Renegade Hi-Flo; Boston Scientific, Marlborough, Massachusetts) was advanced into the uterine artery on both sides (Fig 2). Caudally oriented side-branches of the uterine artery were identified on both sides (Fig 2a, b) and then selectively catheterized. Calibrated microspheres (500–700 μm , 1 mL on each side; Embospheres; Merit Medical Inc., South Jordan, Utah) were administered. On the right side, coiling of a caudally oriented branch was performed (3 mm \times 6 cm and 2 mm \times 4 cm IDC Soft Microcoils; Boston Scientific) before particle embolization to prevent non-target embolization (Fig 2c, arrow). A left-sided branch of the uterine artery was coiled (2 mm \times 4 cm IDC Soft Microcoil; Boston Scientific) after slow microsphere injection (Fig 2d, arrow).

After the procedure, no sign of bleeding occurred within the following 9 weeks. Nonetheless, Doppler ultrasound only showed a slight difference in vascularization before and after the procedure of the embolized region. The cervicoisthmic geminus showed normal growth and normal fetal perfusion determined by Doppler ultrasound measurements. However, the intracavum fetus continued to suffer from progressive placental insufficiency, resulting in an intrauterine demise after 25+5 weeks of pregnancy, with an estimated fetal weight of 245 g.

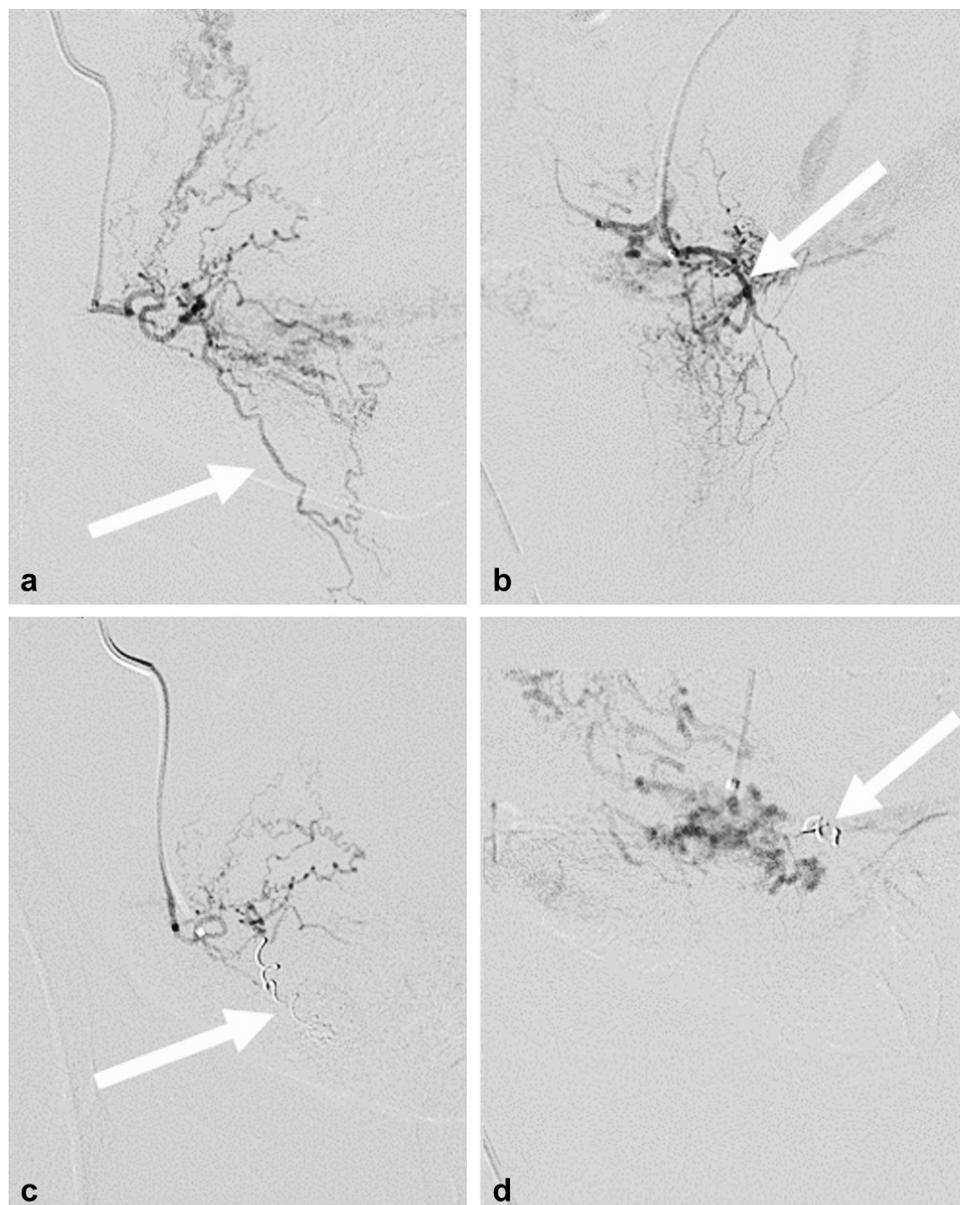


Figure 2. Angiography images showing the placement of catheters and coils during the intervention. The (a) right and (b) left uterine arteries were sequentially catheterized, followed by coil placement on the right (c) and (d) left side. The positions for the coil placement before and after introduction are marked with arrows.

After 28+1 weeks of pregnancy, the patient showed heavy vaginal bleeding. Both children were delivered by cesarean section through fundal uterotomy. The cervicoisthmic AIP was left in situ, and a hysterectomy was performed. The patient received a transfusion of 2 red blood cell concentrates. The birth weight of the living male offspring was 1300 g with a length of 42 cm. The mother was discharged 14 days after giving birth; the newborn after 8 weeks in the hospital.

Follow-Up Examination

Development of the newborn was evaluated after 12 months using the Bayley Scales of Infant and Toddler Development.

Cognitive, language, motor, social-emotional, and adaptive domains were normal, indicating no developmental delays.

DISCUSSION

Cervicoisthmic implantation is defined as a gestational sac localized distally within the cervical channel and the uterine decidua (2). For the present case, it was hypothesized that several curettages resulted in endometrial damage and weakened the internal cervical os (2). An endometrial defect can result in AIP and excessive neovascularization due to uncontrolled and increased trophoblast invasion (5). It has been shown that placental detachment in cervicoisthmic

implantation is associated with life-threatening bleeding requiring hysterectomy (2). Additionally, the placenta expands inside the caudal cavum uteri, explaining severe bleeding during pregnancy as the internal os distends by the growing placenta, and myometrial contractions can cause bleeding by disrupting vessels.

UAE is an efficacious approach in treating ectopic cervical and scar pregnancies in the first trimester (11). UAE's high efficiency in preventing severe bleeding in AIP was recently shown in a study that included 29 cases (12). In this study, no early emergency hysterectomy was necessary, and severe bleeding was prevented, but most patients were at low gestational age or postpartum.

In the presented case, UAE of the cervical branches was performed as an ultima ratio during the 20th week of pregnancy without compromising the fetal wellbeing and resulting in long-lasting cessation of vaginal bleeding. Additionally, a cerclage, leaving the placenta in situ, and a subsequent hysterectomy contributed to the stable course of the pregnancy.

In conclusion, cervicoisthmic implantation commonly results in severe AIP, which usually leads to early hysterectomy and abortion. As part of an individual treatment strategy, UAE prevented early hysterectomy, enabling the live birth of a healthy preterm newborn with subsequent normal development.

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