Research Letter

Interstitial ectopic pregnancy complicated by uterine arteriovenous malformations treated with unilateral transarterial embolization

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Uterine arteriovenous malformation (AVM) often occurs in association with iatrogenic obstetrical procedures such as curettage, cesarean section, and induced delivery related to pregnancy or gestational trophoblastic disease [1]. Interstitial pregnancy complicated by uterine AVM is extremely rare and potentially dangerous. Failure to treat an interstitial pregnancy could result in uterine rupture, massive bleeding, or situations requiring hysterectomy [2]. Herein, we present such a rare case of interstitial ectopic pregnancy complicated by uterine AVM, which was diagnosed by ultrasound and angiography, and treated by unilateral uterine artery embolization.

A 32-year-old woman, gravida 0, para 0, was referred to our hospital with the chief complaint of vaginal bleeding for 2 weeks because of a suspected interstitial ectopic pregnancy. Her vital signs were stable, and a physical examination revealed no rebound tenderness. Her urine beta-human chorionic gonadotropin (β-hCG) test result was positive, and a pelvic examination revealed a mildly enlarged, anteverted uterus, with tenderness in the right fundus on palpation.

Ultrasonographic examination of the patient demonstrated an empty uterus, without a gestational sac. However, an eccentric nonhomogeneous mass measuring 5.56 cm × 5.47 cm in diameter was identified in the right cornual region of the uterus, surrounded by an extremely thin myometrium of <5 mm thick. Furthermore, an echogenic line of endometrium abutted the center of the mass (Fig. 1). Her serum β-hCG level was 807.68 mIU/mL. A right interstitial pregnancy was suspected according to the ultrasonographic findings. The spectral color Doppler imaging showed prominent and chaotic aberrant blood vessels within the cystic lesion (Fig. 2). Computed tomography demonstrated a soft tissue mass located in the right cornual region separated from the endometrial cavity with early venous filling compatible with an AVM.

Three days later, the serum β-hCG level had decreased to 485.90 mIU/mL; thus, the patient was referred for expectant management. In the meantime, the AVM increased to 7.60 cm × 5.63 cm in diameter without internal bleeding or rupture. A color Doppler ultrasonography of the lesion revealed multiple tortuous vessels with continuous blood flow during both systole and diastole phase. The spectral analysis displayed multidirectional, high-velocity, and low-resistance flow within the arteriovenous communications. In addition, resistance index measurements were low, and showed mixing of arterial and venous waveforms. A contrast material-enhanced computed tomography scan through the abdomen showed a well-defined mass-like lesion with internal hypervascularity, early opacified engorged venous structure-associated vascular glomus at the right cornus region of the uterus (Fig. 3). These findings are compatible with uterine AVM.

After receiving counseling on the risk of uterine rupture and possible massive hemorrhage upon rupture, the patient decided to undergo transarterial embolization (TAE). In preparation for TAE, the serial angiography showed a large uterine AVM in the right cornua of the uterus (Fig. 4). The AVM feeding arteries were derived mainly from the right uterine artery and minimally from the distal branch of the left uterine artery. TAE was performed using N-butyl cyanoacrylate glue injected into the right uterine artery. The procedure went smoothly. A post-TAE angiography showed marked obliteration of the AVM nidus and occlusion of the right uterine artery (Fig. 5).

The patient complained of severe lower abdominal pain after TAE, and an oral nonsteroidal anti-inflammatory agent was given for pain relief. The symptoms improved gradually, and the patient was discharged uneventfully on the 2nd day after TAE. One week
later, the transvaginal color Doppler ultrasound showed a persistent cornual mass, but without blood flow inside. The patient’s serum β-hCG level had declined to 3.17 mIU/mL 9 days after the TAE procedure.

In an interstitial ectopic pregnancy, the embryo implants in the interstitial portion of the fallopian tube, accounting for about 2.9% of all ectopic pregnancies [2]. Interstitial pregnancy is often associated with uterine rupture and massive hemorrhage because of delayed diagnosis [3]. In view of the high maternal mortality rate associated with interstitial pregnancy, early diagnosis and treatment are essential to achieve a positive clinical outcome.

Interstitial pregnancy is easily confused with angular pregnancy. Angular pregnancy refers to the implantation located medial to the uterotubal junction in the uterine cavity, and the surrounding myometrial mantle is much thicker. In many cases, the outcome of angular pregnancy is favorable if the layer of myometrium is thicker than 0.5–1 cm and there is no evidence of AVM. The prospect of a successful pregnancy will be good under accurate diagnosis and appropriate management as described by Jansen and Elliott [4]. Interstitial gestation implants outside the uterine cavity, laterally to the round ligament with a thin layer of myometrium [4,5]. On the contrary, ultrasound criteria for the diagnosis of interstitial pregnancy include an empty uterine cavity, eccentrically placed gestational sac surrounded by a thin layer of myometrium that measures <5 mm [5,6]. Moreover, a new four-dimensional volume contrast imaging technology introduced by Chou et al [7] overcomes the limitations of traditional two-dimensional ultrasound imaging, thus offering more accurate and more realistic observations. This method can improve the diagnosis and differential diagnosis between interstitial pregnancy and angular pregnancy or a pregnancy in the anomalous uterus. Missed or delayed diagnoses can occur owing to the rarity of this condition. As a result, uterine rupture occurs in up to 20% of interstitial pregnancies. Our patient had an elevated serum β-hCG level with an intramural hypoechoic cystic lesion in the right cornual region. No intrauterine gestational sac was identified under ultrasonography, which led to the increased suspicion of interstitial pregnancy.

Interstitial pregnancy complicated by uterine AVM is extremely rare and potentially dangerous. Although the cause of AVM is still unknown, it is most often iatrogenic. It usually develops as a result of uterine trauma, such as uterine surgery, use of instrumentation, endometrial carcinoma, and gestational trophoblastic disease [8]. The most common clinical manifestation of uterine AVM is vaginal

Fig. 1. Transvaginal sonography shows a right interstitial pregnancy (large arrow) and endometrium (small arrow).

Fig. 2. Spectral color Doppler imaging of the patient shows high-velocity turbulent flow within the cystic lesion.

Fig. 3. An abdominal contrast material-enhanced computed tomography (CT) scan shows a well-defined mass lesion with internal hypervascularity, early opacified engorged venous structure (large arrows) associated vascular glomus at right cornus region (small arrows).
hemorrhage and menorrhagia [9]. With advances in imaging techniques, transvaginal ultrasonography has become the choice of noninvasive diagnostic testing for detecting AVM [10]. A review article by Peitsidis et al [11] described the most common ultrasound features of AVM including multiple cystic lesions or hypoechoic masses in the myometrium with turbulent blood flow or multiple tortuous feeding vessels. Nonetheless, angiography remains the diagnostic gold standard for AVM diagnosis [9]. The branches of the internal iliac artery, either bilateral or unilateral, typically supply blood to uterine AVMs [11].

There are several different ways to treat AVMs, including ligation of AVM vessels and the uterine artery, local resection of the cornual ectopic mass via laparotomy or laparoscopy, and TAE. Total abdominal hysterectomy is only indicated in women with profuse vaginal bleeding and ruptured AVM or uterus. Because TAE is simple and uncomplicated for surgeons with experience, uterine artery embolization is the most common method of treatment. In the current case, the right uterine artery was embolized because the AVM blood supply was predominantly unilateral, as confirmed by angiography. Varying degrees of pelvic pain and postoperative fever may be observed after embolization [11], and analgesics are frequently used in this condition. The efficacy and safety of TAE for AVM has been demonstrated by several investigators, and both prognosis and outcome are excellent [12–16].

Expectant management of interstitial ectopic pregnancy by regular follow-up is also reported [17,18]. We previously reported a case of cornual pregnancy with spontaneous resolution in which the β-hCG levels returned to the prepregnancy level within 3 months [17]. A high success rate for expectant management is observed in women without signs of internal bleeding and with ectopic masses <5 cm in diameter [18]. We preferred TAE to expectant management for our patient because of the increasing size of the ectopic mass and the high risk of rupture and massive bleeding.

In conclusion, interstitial pregnancy complicated by uterine AVM is very rare. Our patient was successfully treated with unilateral TAE, which is a safe and effective procedure for women desiring future fertility.

Conflicts of interest

The authors do not have any conflicts of interest to declare.

References