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Research Letter

Acquired uterine arteriovenous malformation in a cesarean scar pregnancy

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The recent exponential increase in the rate of cesarean sections has been associated with an increase in placentation complications during subsequent pregnancies [1]. A cesarean scar pregnancy (CSP) is a rare type of ectopic pregnancy that results from faulty implantation within a previous cesarean section scar. CSP may be life-threatening because of subsequent uterine rupture and uncontrolled hemorrhage that require a hysterectomy.

Uterine arteriovenous malformation (AVM), which is an abnormal connection between arteries and veins [2], can be caused by iatrogenic events or pathologic pelvic conditions. With the progress in imaging techniques such as computed tomography (CT) and magnetic resonance angiography, uterine AVM has been reported as one of the complications in CSP cases [3–5]. Although the coexistence of uterine AVM and CSP is potentially life-threatening, optimal management of CSP with uterine AVM has not been determined because it is so rare.

We report a case of CSP complicated by uterine AVM and describe how the patient's fertility was preserved using a combined conservative management approach.

A 32-year-old patient (gravida 3, para 2) was referred for suspicion of a gestational trophoblastic tumor. She had experienced persistent and painless vaginal bleeding for approximately 8 weeks, and ultrasound showed a tumor in the anterior part of the uterus.

The patient had a first cesarean section delivery for breech presentation. She gave birth to a second baby by elective cesarean section 2 years prior to presentation for the current problem. This pregnancy was followed by one missed abortion during the first trimester, for which she underwent uterine evacuation.

On pelvic examination, moderate vaginal bleeding and a slightly enlarged uterus were observed. Transvaginal ultrasound revealed a tumor of 60 mm × 45 mm in size with an unclear boundary on the previous cesarean section scar site. The uterine cavity was empty and no fluid was found in the cul-de sac. Color Doppler ultrasound showed numerous tortuous blood vessels encompassing the tumor (Fig. 1).

The patient's serum β-hCG level was 38,978 IU/L. As no evidence of a clear gestation sac or embryo was found in the mass, biopsy via uterine curettage was necessary for a definitive diagnosis. Considering the highly vascularized tumor and persistent vaginal bleeding, uterine artery embolization (UAE) was first performed. A pelvic angiogram during UAE showed that the mass was surrounded by multiple feeders from internal iliac arteries (Fig. 2). Uterine curettage was successfully performed without abnormal bleeding, and histopathology revealed necrotic placenta remnants and a conception product.

After this procedure, we monitored the patient weekly in terms of serum β-hCG and transvaginal ultrasound. Her serum β-hCG level plateaued after it had decreased to 1005 IU/L, and a remaining mass with blood flow was still evident on transvaginal ultrasound scans. Therefore, she was treated with

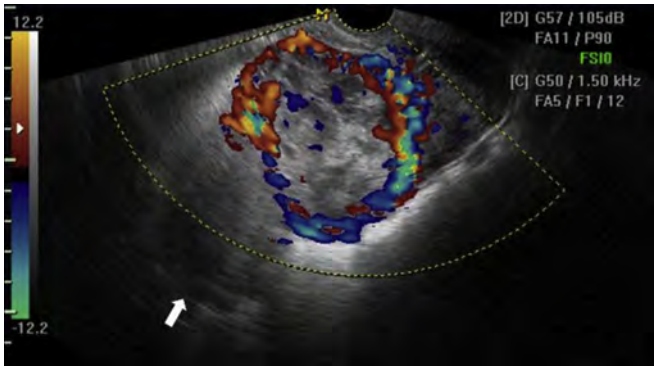

Fig. 1. Color Doppler ultrasound showing numerous tortuous blood vessels encompassing the tumor in the anterior isthmic region of the uterus (arrow, uterine fundus).

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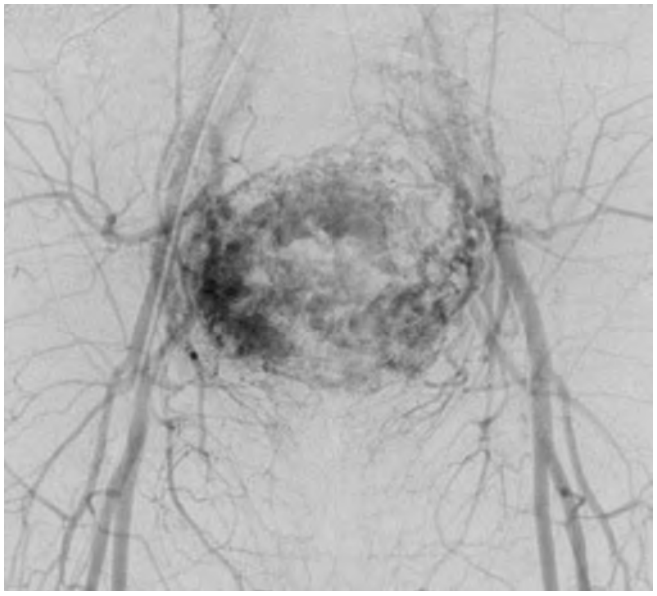

Fig. 2. Pelvic angiogram showing uterine arteriovenous malformation supplied by multiple feeders from branches of both internal iliac arteries at the initial uterine artery embolization.

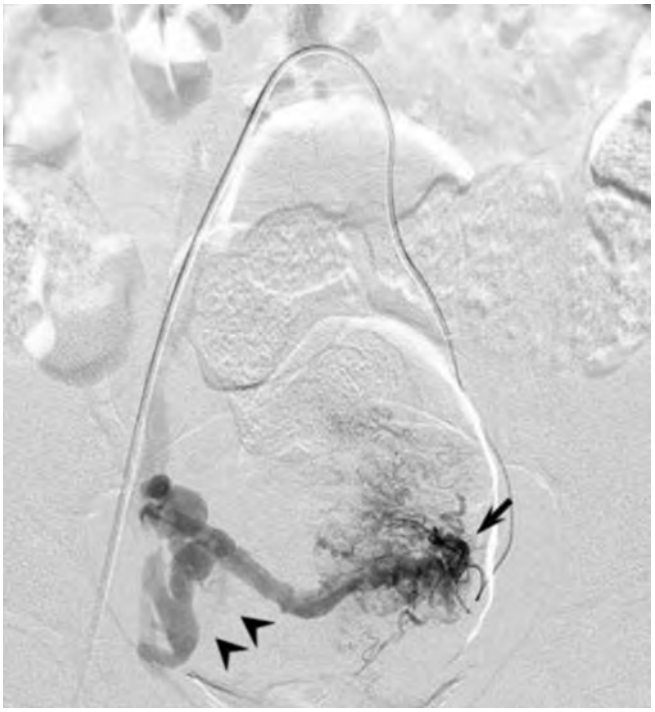

Fig. 3. During repeat uterine embolization, a pelvic angiogram of the left uterine artery (arrow) revealed simultaneous filling of veins (arrow head) at the arterial phase, which is a definitive diagnostic finding in arteriovenous malformation.

multidose systemic methotrexate (MTX) and folinic acid on alternate days. After three courses, the patient's serum β-hCG decreased to <5 IU/L.

Approximately 4 weeks later, the patient visited the emergency department with massive vaginal bleeding and her vital signs were unstable. A CT scan showed a suspicious transverse vascular malformation in the lower uterine segment. After the patient and her husband both expressed a desire to preserve her fertility, the decision was made to manage her condition conservatively with UAE for emergent hemostasis. During selective UAE, an angiogram of the left uterine artery showed left aneurismal dilation, and AVM was observed in the right uterine vein (Fig. 3). After selective UAE on both sides, the vaginal bleeding lessened. Then uterine curettage was performed to remove the remaining placenta tissue. The patient resumed menstruation 3 months after curettage. In CT images obtained 7 months postoperatively, no findings of remnants or abnormal blood vessels were observed at the previous cesarean scar site. The patient reported a regular and normal menstruation cycle.

CSP, a rare type of ectopic pregnancy, is caused by blastocyst implantation at the scar site for a previous cesarean section. Scar tissue surrounding the gestational sac in CSP may increase the susceptibility to placenta-associated complications. Uterine AVM involves abnormal communication between intramural branches of the uterine artery and the myometrial venous plexus [2]. It can be caused by the inherently erosive nature of synthiotrophoblastic tissue and chorionic villi during placental formation in CSP, in which the defective decidual layer induces the development of abnormal connections among vascular structures.

Most uterine AVM cases are congenital, but there have been reports of acquired cases associated with previous uterine surgical procedures or gestational trophoblastic disease [2]. Traumatic causes of arterial-to-venous connections include endometrial curettage, cesarean section, and pelvic surgery. Pelvic examination may reveal transcervical bleeding or a palpable pulsating mass accompanied by lower abdominal pain, dyspareunia, and anemia. Endometrial curettage performed in patients with dysfunctional uterine bleeding worsens vaginal hemorrhage by uterine AVM [6]. In the past, uterine AVM was usually diagnosed through pathology examinations after hysterectomy. At present, Doppler ultrasound, hysteroscopy, CT scanning, and magnetic resonance imaging are used as diagnostic modalities, and a definite confirmation can be made via angiography. Early venous drainage into the endometrial or myometrial veins is a sign of arteriovenous shunting observed in AVM [7].

Uterine AVM in CSP may be a natural response of the inherently erosive trophoblastic tissue as it establishes an adequate blood supply for the placenta. Restriction due to the fibrous tissues and defective endometrium of cesarean scars might promote abnormal angiogenesis. We incidentally detected uterine AVM while performing a second UAE to control massive hemorrhaging. During the first UAE, there was no definitive evidence of AVM, but numerous tortuous vascular components surrounded the tumor. A literature review revealed three cases of CSP-associated uterine AVM [3–5]. A definitive diagnosis was difficult in the past because of limited vascular imaging modalities. However, more cases of CSP-associated AVM have been reported

owing to recent increases in UAE as an adjuvant treatment for CSP.

The diagnosis of CSP has recently increased because of the common use of early pregnancy transvaginal ultrasound and the higher rate of cesarean delivery. Management for CSP, however, is varied, and there has been difficulty in establishing an optimal treatment. Medical treatment involves systemic or local administration of MTX, injections of embryocides or oral uterotonics such as mifepristone, and varying combined regimens [8]. Uterine curettage or hysterectomy has been occasionally used as a surgical approach. It has been reported that aspiration of the ancillary surgical sac to prevent scar rupture during medical treatments is effective in reducing the size of the conception product [9]. Salvage treatments, such as laparoscopic or hysteroscopic resection of the lesion, have recently been used to preserve the uterus [10,11].

As an additional treatment, selective UAE has been considered for patients who want to retain future fertility. Risks include uterine infarction or ischemia and necrosis; however, the majority of patients tolerate the UAE procedure well [12,13]. Recurrent bleeding after UAE may develop, depending on the presence of multiple arterial feeders or the embolization medium, such as gelatin foam particles [5,14]. Despite these side effects, UAE is a minimally invasive procedure under local anesthesia or intravenous sedation that is associated with lower morbidity, shorter hospitalization, and uterine preservation [2]. In this case, UAE was performed twice during conservative management. The first UAE was performed as a prophylactic procedure to avoid massive uterine bleeding during curettage for tissue biopsy. The repeat embolization performed to stop massive bleeding revealed the uterine AVM, which was successfully treated.

Uterine curettage is not recommended as a first-line therapy for CSP, as it has either been unsuccessful or has caused complications, including an increased risk of uterine rupture and resulting secondary bleeding requiring hysterectomy, because of the abnormally increased vascularity and difficult surgical approach [8]. In addition, as the gestational sac in CSP is not located in the endometrial cavity, uterine curettage may be incomplete and leave remnants of invasive trophoblastic tissue, which increases the risk of associated morbidity.

Histopathologically, placental implantation in CSP is similar to placenta accreta. The defective decidual layer of the previous scar seems to be susceptible to faulty invasiveness of the gestational product in CSP. In the case reported here, a biopsy via uterine curettage was required for differential diagnosis from gestational trophoblastic disease because no definite finding of a conception-like gestation sac, embryo, or double-ring sign was observed in the tumor. Although we do not presume that conservative management of CSP is a direct cause of AVM in this case, incomplete uterine curettage and remaining trophoblastic tissue may partly contribute to AVM occurrence. We suspect that the remaining placental tissue exhibited neovascularization and that the uterine AVM was caused by an intrinsic attribute of the chorionic villi. In addition, uterine curettage may be a traumatic factor in causing artery-to-vein connections.

Our case of CSP complicated by uterine AVM was managed with combined conservative treatment involving systemic MTX, uterine curettage, and UAE, which preserved the patient's fertility. We propose that UAE should be considered in CSP management for cases with numerous tortuous vascularities. If remaining placental tissue is expected after uterine curettage during conservative management of CSP, clinicians should consider the development of an uncommon fatal complication, such as uterine AVM, despite resolution of serum β-hCG.

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