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Uterine arteriovenous malformation or uterine artery pseudoaneurysm secondary to uterine aspiration in cesarean scar ectopic pregnancy: a case report and review of the literature

Ngoc Diep Le[1†] and Phuc Nhon Nguyen[2,3,4,5*†]

Abstract

Introduction Uterine arteriovenous malformation or uterine artery pseudoaneurysm after the treatment of cesarean scar ectopic pregnancy is a scarce entity, leading to potentially life-threatening conditions due to the massive bleeding. The management remains significantly crucial. Herein, we report an uncommon case with surgical management of such a rare condition at our center.

Presentation case A 31-year-old Vietnamese female patient (gravida 2, para 1) was hospitalized for cesarean scar pregnancy. The uterine aspiration was well performed. However, the woman was readmitted for an abnormally persistent hypervascularity at the site of cesarean scar ectopic pregnancy under repeated ultrasound scans. After counseling, arteriovenous malformation was suspected more, whereas uterine pseudoaneurysm was incompletely ruled out. Initially, gonadotropin-releasing hormone agonist was administered, with two doses of 37.5 mg. Later, the patient underwent laparoscopic intervention to suture the vascular proliferation mass to prevent spontaneous rupture. The surgery was achieved successfully. One more dose of gonadotropin-releasing hormone agonist was added. The ultrasound detected no existing lesion compared with previous ultrasonic imaging and serum beta-human chorionic gonadotropin declined to a negative value. The patient was monitored in 1 month uneventfully.

Conclusion A high index of suspicion of uterine arteriovenous malformation or uterine artery pseudoaneurysm should be raised after uterine curettage of cesarean scar ectopic pregnancy. Ultrasound scan is still a pivotal first-line tool in assessing this abnormality. Laparoscopic surgery with compressing suture of the enhanced myometrial vascularity/arteriovenous malformation combined with administration of gonadotropin-releasing hormone agonist may be applied. This surgical approach has not been reported before, which makes our case report unique. Further cases are required for this rare entity so as to ensure patient safety.

[†]Ngoc Diep Le and Phuc Nhon Nguyen have contributed equally to this work and share the first authorship.

*Correspondence:
Phuc Nhon Nguyen
docternhon@gmail.com; phucnhon.nguyen@chu-orleans.fr
Full list of author information is available at the end of the article

Key Points

- A suspicion of uterine arteriovenous malformation or uterine artery pseudoaneurysm could be present as an unexpected complication after uterine curettage of cesarean scar ectopic pregnancy.
- Ultrasound scan remains an initial tool in assessing the enhanced myometrial vascularity.
- Laparoscopic surgery with compressing suture of the arteriovenous malformation combined with administration of gonadotropin-releasing hormone agonists may be considered in treatment.

Keywords Arteriovenous malformation, β-hCG, Cesarean ectopic pregnancy, Massive bleeding, Laparoscopy, Pseudoaneurysm, GnRH-a, Ultrasound, Uterine curettage, Vietnam

Introduction

Cesarean scar ectopic pregnancy (CSEP) is defined as full or partial implantation of the gestational sac in the scar of a previous cesarean section [1]. Classically, the classification of CSEP depending on the location of the gestational sac consists of type 1 (on the scar) and type 2 (in the niche) [2]. The incidence of CSEP has been estimated to range from 1/1800–1/2500 of all previous hysterotomies [3]. The risks of CSEP are active bleeding, miscarriage, rupture, abnormally invasive adhesion of the placenta, and hysterectomy [4–7].

Nevertheless, arteriovenous malformation (AVM) or uterine artery pseudoaneurysm (UAP) is an extreme rarity. AVM is a pathologic phenomenon described as a faulty "short circuit" or bypassing capillary network of the bloodstream between an organ's arterial and venous supply [8]. It could develop elsewhere in the body, such as the brain [9]. Uterine AVM is a rare entity in gynecology, with fewer than 100 cases reported in the literature since Dubreuil and Loubat reported the first case of AVM in 1926 [10].

Ultrasound (US) demonstrates turbulent arterial flow with a to-and-fro or Yin-and-Yang pattern that results from blood flow into the pseudoaneurysm. Histologically, UAPs often consist of only one layer of loose connective tissue, distinguishing them from true aneurysms consisting of a complete three-layered wall. Extraluminal turbulent blood flow can lead to enlargement of the UAP, making it susceptible to rupture and subsequent bleeding. Two types of UAPs have been reported: one that communicates with the uterine cavity and causes genital bleeding and one that has no communication and results in hematoma formation outside the uterus [11]. The underlying mechanism is still unclear. These vascular abnormalities occur after various traumatic obstetric or gynecological procedures, but also occur secondary to non-traumatic etiologies [11–13].

Uterine AVM may be congenital or acquired. However, this condition could be rarely detected following surgical intervention on the uterus such as cesarean section, uterine dilation, and curettage [8]. The AVM as a consequence of CSEP has been documented recently due to the increasing rate of cesarean delivery and advances in imaging modalities [14, 15]. According to I. E. Timor-Tritsch, a retained product of conception (RPOC) may be associated with enhanced myometrial vascularity [16].

Uterine AVM especially could be mimicked with pseudoaneurysm, gestational trophoblastic disease (GTD), uterine pseudoaneurysms, hemangiomas, varicosities, and malignancies of the uterus such as sarcomas

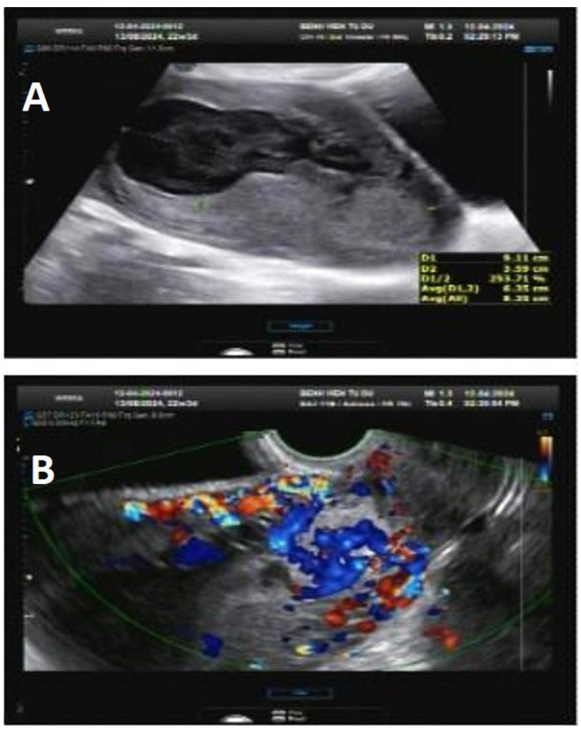

Fig. 1 Sagittal view of the uterus via transvaginal ultrasound shows numerous hypoechoic regions at the previous cesarean section site, extending to a fundal uterine cavity (**A**). Using color Doppler transvaginal ultrasound, cesarean scar ectopic pregnancy was diagnosed, but partial molar pregnancy could not be excluded because of a hypervascular mass (**B**)

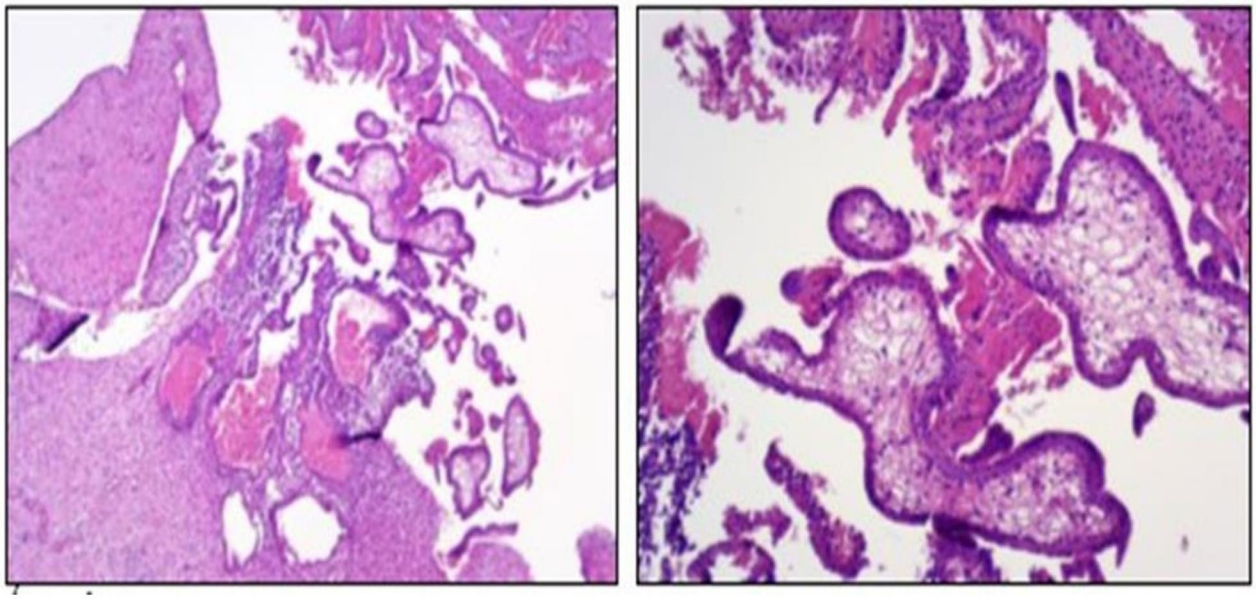

Fig. 2 Hematoxylin and eosin stain showing the trophoblastic tissue corresponding to gestational structure (×10 **A**, ×40 **B**)

[17]. The clinical presentation is associated with severe hemorrhage if the management is delayed. Imaging modalities could be made by ultrasonography, magnetic resonance imaging (MRI), computed tomography angiography (CTA), and digital subtraction angiography (DSA) [13, 17]. Ultrasound scan plays an important role as the primary imaging tool in detecting the abnormal vascular proliferations [18]. On color Doppler, tortuous vascular structures with multidirectional shunt flow, low resistance index, and high velocity are highly suggestive of arteriovenous malformation [19].

Accordingly, the multimodal treatment for enhanced myometrial vascularity should be individualized depending on the patient's desire for future fertility and their hemodynamic stability [16]. Previously, expectant management and medical treatment could be encountered. During postpartum, several UAP cases may resolve spontaneously, without further surgical intervention [20]. Progestins and gonadotropin-releasing hormone agonists (GnRH-a) have been the most studied medical therapies and were efficacious with low complication rates [21]. Uterine artery embolization (UAE) has been widely reported for conservative management, but its cost remains high in low-resource settings [22, 23]. Moreover, UAE has been associated with infertility, premature ovarian failure, and uterine synechia [21]. Ultimately, fertility-sparing surgery could be an alternative option to hysterectomy in selected patients [24].

In this article, we shed light on a rare case of suspected uterine AVM as the consequence of CSEP at our tertiary referral hospital. The patient underwent laparoscopy with compressing suture of the AVM mass with a successful outcome. To our best knowledge, a similar case with this surgical approach has not been reported previously. In addition, we conducted a literature review to enhance the comprehension concerning this rare entity.

Presentation case

A Vietnamese female patient aged 31 years with gravida 2 and para 1 was hospitalized due to suspected pregnancy. The patient had one cesarean delivery without a psychosocial history. She denied consuming alcohol, tobacco, or cannabis. The history of her family was unremarkable. Her vital signs were stable. She complained of a delayed menstrual period and slight vaginal bleeding without abdominal pain. On gynecologic examination, her uterine size corresponded to the gestation of 7 weeks. On per speculum examination, the cervix was completely normal. A beta-human chorionic gonadotropin (β-hCG) level of 58,644 mUI/mL confirmed pregnancy. Ultrasound scan detected an empty uterine cavity and an irregular gestational sac without an embryo located near the previous cesarean scar site about 20 mm. However, the CSEP diagnosis was carefully differentiated with partial hydatiform mole due to vascular proliferation visable upon Doppler ultrasound (Fig. 1A, B). The patient underwent a uterine aspiration under ultrasound guidance in

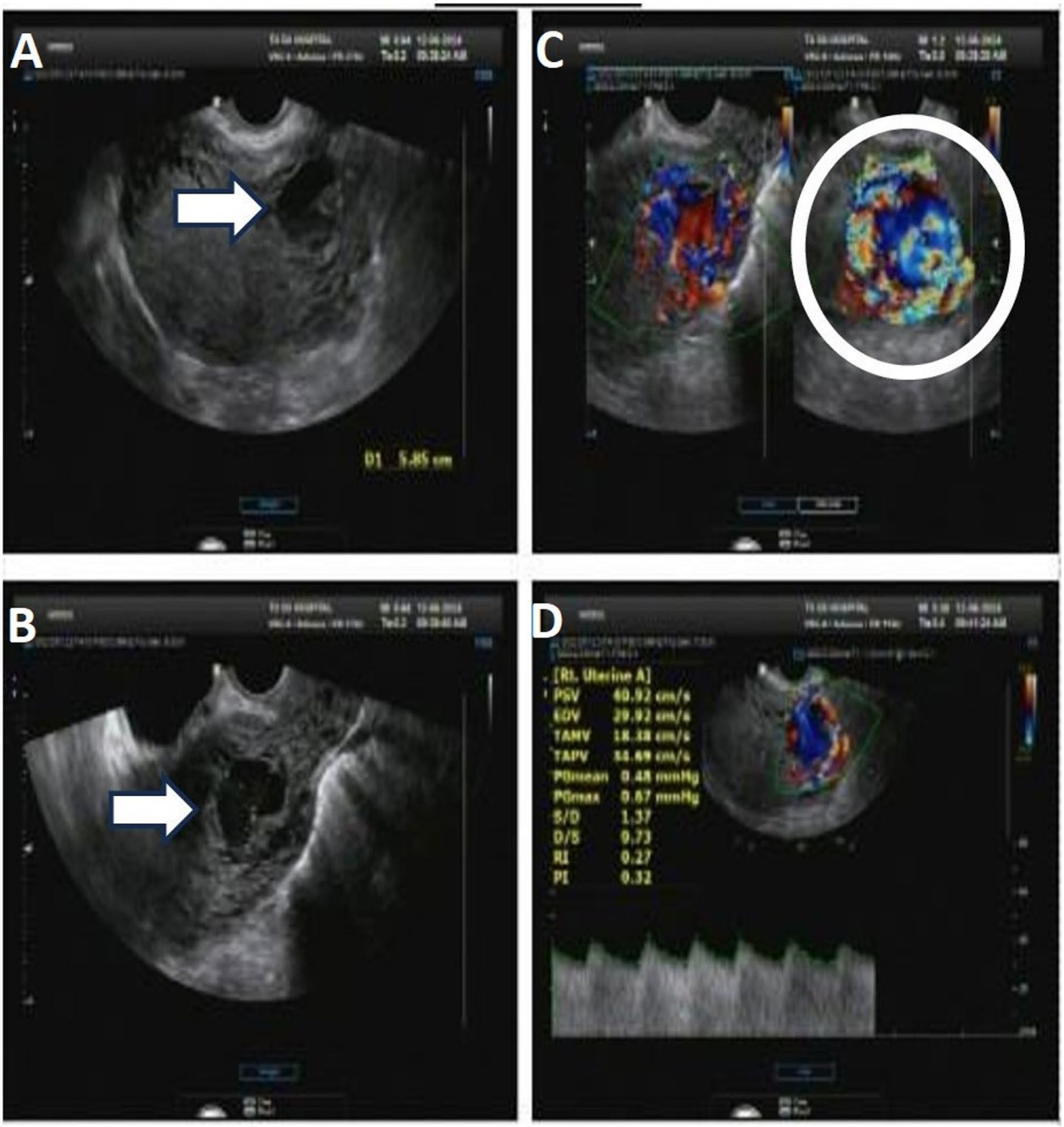

Fig. 3 An anechoic complex cystic structure containing multiple tubular serpentine cystic spaces in the lower anterior wall of the uterus close to the site of the previous cesarean scar of the lower uterine segment (white arrow). Hypervascular mass located at the cesarean section site, presumably representative of cesarean scar pregnancy. The endometrial lining was thin with no signs of intrauterine or extrauterine gestation (**A**–**C**). Color Doppler ultrasound sagittal view of the uterus shows tortuous structure with mixed arteriovenous flow, color aliasing, and flow reversal as well as an increased peak systolic velocity of 40.92 cm/second, and a low resistive index of 0.27, indicating possible arteriovenous malformation (white circle) (**D**)

the operating room. After procedure, 70 g of products of conception were sent for histopathological examination (Fig. 2A, B). The total blood loss was 20 mL. Her β-hCG level declined to 32,240 mUI/mL after 24 hours.

The patient was monitored on ultrasound every 2 weeks with an abnormal hypervascularity (Fig. 3A–D). Initially, the woman received also two doses of gonadotropin-releasing hormone agonists (GnRH-a). A total of 3

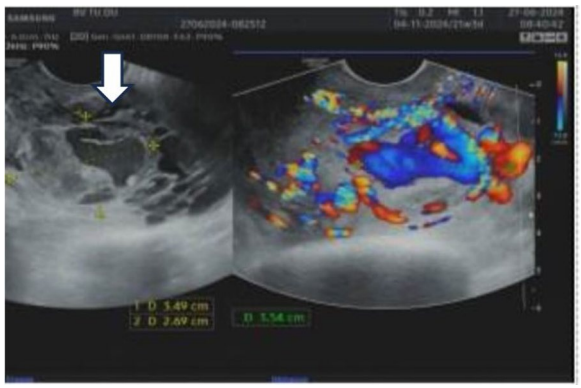

Fig. 4 Gray-scale ultrasound image (black and white) reveals irregular, inhomogeneous, hypoechogenic, tortuous structures within the myometrium (white arrow) (**A**). Dilated vessels with a corkscrew appearance, resembling varicose veins with interconnecting fistula. A tortuous network of dilated vascular channels with turbulent flow was observed (**B**)

months later, the patient was readmitted due to an abnormally proliferative vascular structure at the previous CSEP site. She was asymptomatic and hemodynamically stable, except for a small amount of brown discharge. Her β-hCG level was measured at 14.59 mUI/mL. Serum laboratory tests were completely normal. However, the mass progressed rapidly. Ultrasound scan revealed a hypervascular mass measuring of 33×16×29 mm with the highest peak systolic velocity (PSV) in the center of the "lake" of 41.4 cm/second and a resistance index (RI) of 1.0 (Fig. 4A, B). The residual myometrial thickness at the previous cesarean site was thin. Arteriovenous malformation was suspected, differentiating from pseudoaneurysm.

After consultation, the patient underwent laparoscopy with ultrasound guidance with compressing suture of the aneurysm. Upon laparoscopy, a slightly blue–purple mass measuring 5×5 cm in size was found at the left side of the uterine isthmus (Fig. 5A–D and Supplemental Video 1). A compressing suture of the abnormal mass and bilateral uterine artery ligation was carefully performed (Fig. 6A–D; Supplemental Video 2). In addition, a hysteroscopy was performed, assuring an empty uterine cavity (Supplemental Video 3). The ultrasound immediately confirmed the loss of the abnormally proliferative vascular structure after surgical intervention at the end of the laparoscopic procedure in operating room (Fig. 7A, B). The total blood loss was 200 mL, and no severe bleeding was noted. After surgery, the patient received one more dose of GnRH-a administration. Additionally, broad-spectrum antibiotic therapy was given to prevent the postoperative infection. Her menstruation resumed 1 month after surgery. After surgical intervention, the patient was monitored without complication during the 8-month post-surgery follow-up visit. The menstruation was regular. Neither abnormal vaginal bleeding nor abdominal pain was noted. The ultrasound control confirmed the complete disappearance of the AVM. To ensure mental well-being and physical health, pregnancy had not yet been planned 1 year after the intervention. A subsequent pregnancy would be still monitored in the next years. The patient and her family were deeply grateful to the team for treating her challenging case.

Discussion

In the present case, the diagnosis of CSEP was clearly identified by the criteria on ultrasound and confirmed by histopathological examination. The hydatiform molar pregnancy was completely excluded. Moreover, after uterine aspiration, the β-hCG concentration declined rapidly. The management with ultrasound-guided uterine aspiration was appropriate. This approach was recommended by the Society for Maternal–Fetal Medicine (SMFM) [25]. The surgical procedure was completed unremarkably. However, on ultrasonic monitoring, the patient had a suspected uterine hypervascularity during 2 months. The dilemma diagnosis was usually made between uterine pseudoaneurysm and arteriovenous malformation on the basis of predisposing risk factors of CSEP.

Clinically, abnormal vaginal bleeding and abdominal pain could be present among cases with enhanced myometrial vascularity. Vaginal bleeding is the most common symptom of uterine AVM, varying from light spotting to heavy hemorrhage (Table 2). Rarely, a pulsatile mass may be palpable during digital examination [11]. Uterine arteriovenous malformations (AVM) are vascular disorders characterized by complex high-flow tangles of abnormal vessels connecting arteries and veins with bypassing capillaries. Most AVMs are acquired. The term "enhanced myometrial vascularity" (EMV) describes any condition in which any uterine pathology may lead to increased myometrial vascularity regardless of the absence or presence of residual tissue of gestation [26]. During the postpartum period, AVMs should also be differentiated from subinvolution of the placental bed, and postpartum uterine pseudoaneurysms. Subinvolution of the placental bed presents as hypervascular lesions that do not have fistulous connections between arteries and veins. Recently, a rare case of AVM after a full-term vaginal delivery with massive blood loss of 1000 mL has been documented [27]. In contrast to AVM, pseudoaneurysm is a saccular dilatation of the uterine artery, without communication with the venous system [17]. The severity of the pseudoaneurysm depends on the fragility of its wall. Gray-scale ultrasound is mostly nonspecific, detecting an anechoic

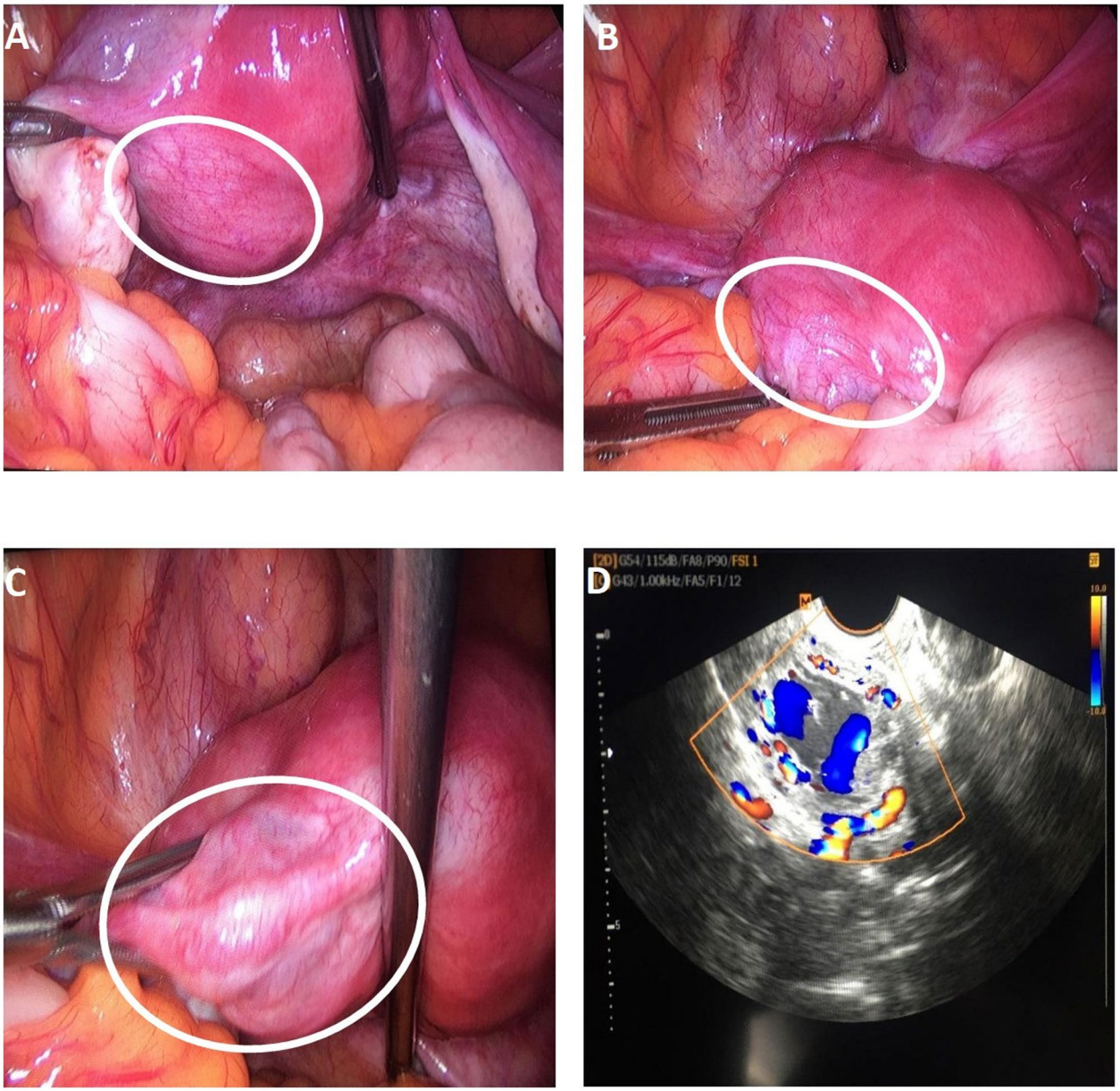

Fig. 5 Upon laparoscopy, an abnormally protruding bluish mass measuring about 5 × 5 cm was observed macroscopically at the left uterine ithmus (white circle) (**A**–**C**). A color Doppler ultrasound was added to determine the location of the arteriovenous malformation mass (**D**)

mass within the uterine myometrium. Doppler ultrasound imaging showed swirling blood flow [11]. Nevertheless, the occurrence of this structured abnormality remains not well known.

Histopathologically, AVMs may be classified as cirsoid or cavernous depending on the number and diameter of the intralesional vessels [17]. Histopathological examination as the hallmark criterion could identify the AVM by showing numerous large ectatic vessels within the myometrium [28]. Conversely, different from the congenital form, acquired uterine AVMs are more common and are characterized by abnormal communication between uterine arteries and the myometrial venous plexus, deep within the myometrium and endometrium [17]. Cirsoid AVMs have multiple, dilated vessels with a corkscrew appearance, resembling varicose veins with interconnecting fistula. The cavernous type includes a single arterial vessel feeding many small connecting fistulas [17].

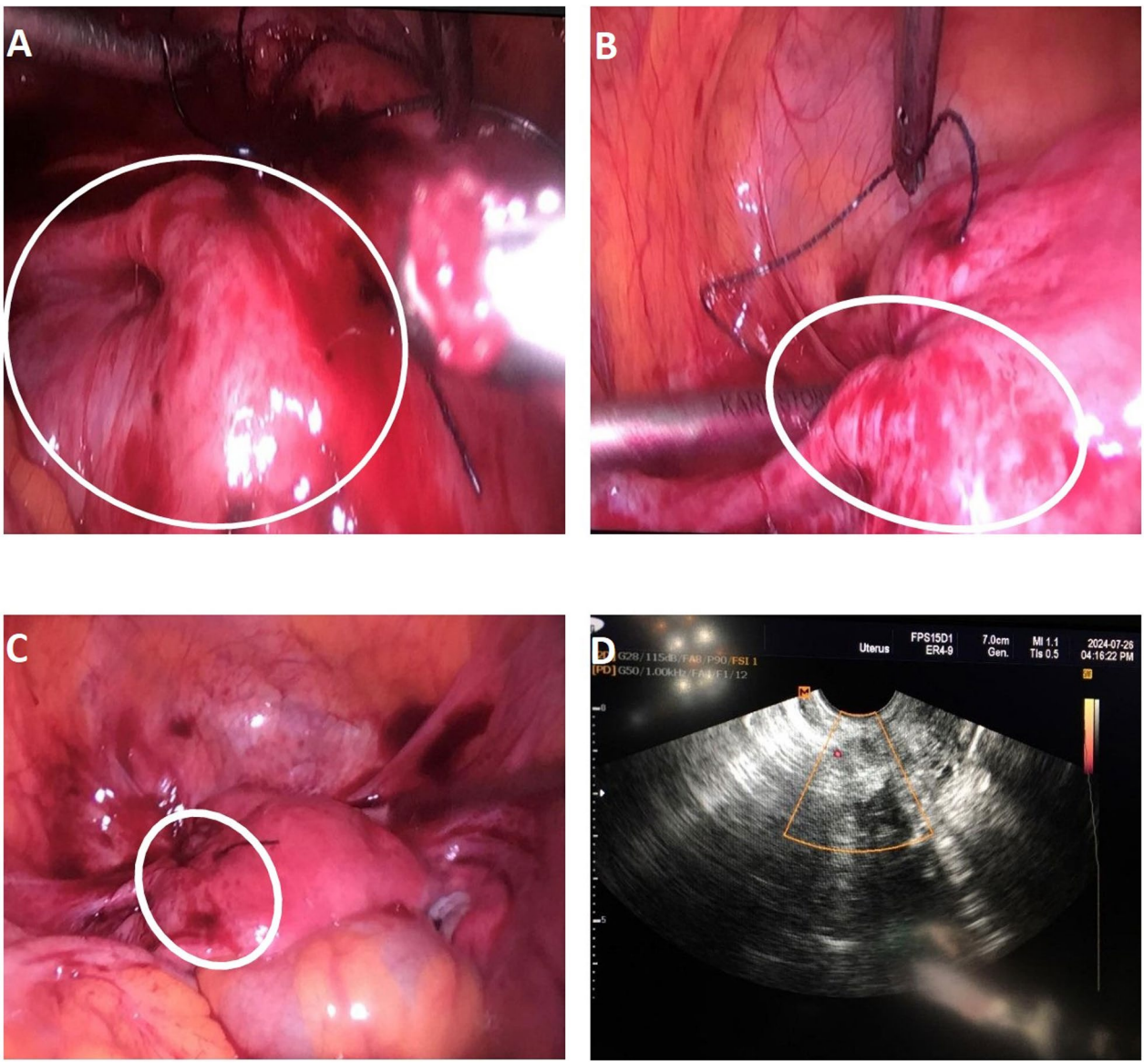

Fig. 6 The surgical intervention with compressing suture and bilateral uterine artery ligation was made meticulously. The protruding mass (white circle) completely disappeared after the procedure (**A**–**C**). Turbulent flow was absent on the Doppler ultrasound (**D**)

In our case, acquired AVM following uterine aspiration and the cirsoid form was suspected, following the diagnostic flowchart of Giurazza *et al.* [13]. Overall, two-dimensional color Doppler ultrasonography cannot reveal the precise vascular structure of the offending vessel from which the pseudoaneurysm has developed so as to determine the management [11], whereas a three-dimensional color Doppler angiogram reveals the configuration of the enhanced myometrial vascularity/arteriovenous malformation and its main feeding and draining vessel [8]. On spectral Doppler ultrasound, multiple vessel patterns should be differentiated from endometrial cancer and other gestational trophoblastic diseases [29, 30]. According to several studies, accurate imaging assessment for AVM was an angiogram [17, 18].

According to Hoang *et al.*, ultrasound is used for the initial estimation of uterine AVM. Ultrasound reveals intense vascularity with the chaotic and omnidirectional flow. Lesions have high peak velocity and low resistance flow on spectral Doppler. Computed tomography (CT) and MRI are noninvasive methods that provide valuable compatibility with digital subtraction angiography (DSA) to support the diagnosis and treatment of uterine AVM. CT concisely shows multiple dilated feeding arteries,

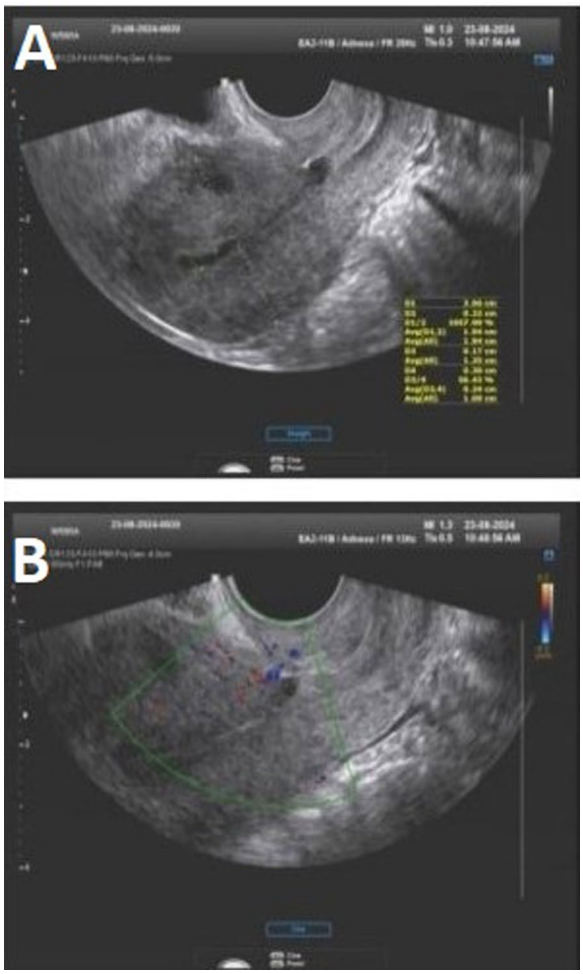

Fig. 7 The mass reduced in size, and the vascularity disappeared gradually on the ultrasonic follow-up in the second week after surgical intervention (**A**) and after 1 month (**B**)

enlarged draining veins, and central nidus. Meanwhile, MRI findings comprise a bulky uterus, vague mass, and focal or diffuse disruption of the junctional zone. The presence of lesions of multiple circuitous and serpiginous flow related signal voids in the myometrium and parametrium. DSA reveals immediate opacification, filling of many enlarged veins from a nidus fed by bilateral hypertrophic uterine arteries [31]. Nevertheless, due to the limitation of low-resource settings and the risk of ruptured mass, the angiography was initially ignored in the present case. In terms of MRI, a prominent lesion with multiple serpentine-like pathways suggestive of aberrant vessels in the wall of the uterine body was reported [27].

Both uterine AVM and uterine artery pseudoaneurysms required a timely, accurate diagnosis and a proper treatment for better prognosis. Regardless of the chosen management, some appropriate modalities should be based on the patient's condition, the experience of the surgeon, and available resources [21]. The clinical decision-making in AVM patients should be based mainly on imaging of the features of vascular proliferation instead of β-hCG levels since β-hCG levels remain persistently low after CSP treatment, as in our case and in the previous cases (Tables 1, 2). According to Wu *et al.*, the location of the mass may be more predictive in prognosis than its size [11]. Previously, endometrial curettage can catastrophically worsen bleeding from this vascular malformation [28]. Hysterectomy should be quickly made if the bleeding is torrential [32, 33]. A normal myometrium shows a peak systolic velocity (PSV) of 9–44 cm/second and resistive index (RI) of 0.6–08, whereas AVM will show high velocity, mean PSV (136 cm/second) and low resistance flow (mean RI, 0.3), along with low pulsatility of the arterial waveform and PSV consistent with an arterial flow pattern of venous flow [10]. In line with the findings of Timor-Tritsch *et al.*, based on PSV of the AVM shunt, cases between 40 and 60 cm/second may be managed expectantly, provided they do not continue with prolonged or severe bleeding. With a PSV of more than 60–70 cm/second, however, UAE may be considered as the treatment of choice [8]. Accordingly, conservative medical management could be chosen if the mass is small and the PSV < 60 cm/second. If conservative management fails or the mass is large with a PSV > 60 cm/second, embolization could be replaced. Hysterectomy should be discussed in difficult cases (Table 2).

Alternatively, an emergency endovascular intervention with selective uterine artery embolization may be performed in recent years [10, 17, 34–40]. In the report of Hammad *et al.*, embolization of the arteriovenous malformation was performed successfully in three cases, and one case was managed conservatively on hormones. After treatment, two of them conceived within a year and had live births at term [18]. In the largest systematic review on 371 patients, Labarta *et al.* found that the global success rate after embolization treatment was 88.4%, presenting a low risk of adverse outcomes (1.8%), even in women with later pregnancy (77% had no complications) [41]. However, selective UAE may fail because of the thin and curved blood vessels [42]. Additionally, recurrent massive vaginal bleeding after UAE could be occurred [24]. In available resources, bilateral uterine artery embolization could help stop severe massive bleeding in emergent conditions and prepare for delayed hysterectomy [43]. Rarely, Kim *et al.* have documented a case wherein a patient underwent hysteroscopic technique for AMV excision. The study suggested that hysteroscopy could be prioritized in the management of uterine AVMs in women of reproductive age. However, an AMV mass should be present in the endometrial cavity, with a small size [44].

Table 1 The characteristics of the present case following the time points of treatment

Time points	US findings	β-hCG (mUI/mL)	Management
First hospitalization	-Irregular gestational sac measuring 19 × 12 × 18 mm without an embryo located about 20 mm from the previous CS site -Myometrial thickness at CS site of 3.1 mm -Subchorial fluid 54 × 30 × 50 mm -CSP classified as COS-2 and type I -Differentiated image with trophoblastic disease due to vascular proliferation	58,644	After two monitoring US in 21 days, intervention by uterine aspiration with US guide for suspicion of CSP was performed.
24 hours after first intervention	Fluid collection in uterine cavity without abnormal vascular proliferation	32,240	Additional MTX (two doses)
-After 2 months of uterine aspiration -Outpatient treatment	Presence of abnormal vascular proliferation	54.58	Administration of GnRH-a (the first dose).
-After 2.5 months of uterine aspiration -Outpatient treatment	Suspicion of AVM or pseudoaneurysm after treatment of CSP	36.99	Administration of GnRH-a (the second dose).
-After 3 months of uterine aspiration -Second hospitalization	Rapid evolution of the abnormal proliferative mass in the uterine cavity	14.59	Surgical intervention.
After 1 month of surgical intervention	No intrauterine vascularity on US	< 5.00	Follow-up.

AMV, arteriovenous malformation; β-hCG, beta-human chorionic gonadotropin; CS, cesarean section; CSEP, cesarean scar ectopic pregnancy; MTX, methotrexate; GnRH-a, gonadotropin-releasing hormone agonist; US, ultrasound

In our case, in addition to the medical treatment using GnRH-a, the woman underwent laparoscopy for compressed suture of the abnormal proliferative vascular mass along with bilateral uterine artery ligation. Laparoscopy as a minimally invasive procedure has been described as an alternative to laparotomy for its cosmetic advantage and shorter recovery time. During surgery, conversion to laparotomy could be replaced with laparoscopy if bleeding is uncontrollable. In this case, AVM excision was not chosen since the mass size presented a worsening, high risk of severe hemorrhage. To obviate the recurrence of AVM, a repeated ultrasound should be performed, and the retained product of conception should be completely ruled-out [8, 45]. In the present case, menstruation was observed after 1 month of surgical intervention. Due to the uncommon occurrence of this pathology as well as the paucity of data concerning our technique in literature, the long term effects relating to subsequent pregnancies and uterine integrity have not been well known. In future pregnancy, some complications include uterine infection, uterine ischemia, uterine necrosis, and damage to other organs, and abnormalities of the placenta should be noted [31]. Regarding the laparoscopic intervention, Ciebiera *et al*. reported that laparoscopy could close the feeding vessel or remove the pathological tissue in the management approach of uterine artery pseudoaneurysm. This method solves the problem permanently and could be applied in other centers after a successful case series with long-term reproductive outcomes [46].

Conclusion

In summary, enhanced myometrial vascularity resulting in AVM or pseudoaneurysm ought to be taken into account after surgical intervention for CSEP. Ultrasound scan remains a mandatory first-line modality for follow-up in low-resource settings. In our case, compressing suture with bilateral uterine artery ligation combined with administration of GnRH-a may be acceptable for this uncommon pathology. Since the technique has not been documented previously, further data are necessary for elucidating a such choice as well as evaluating the efficacy, safety, and impact on future fertility.

Table 2 Summary of cases relating to arteriovenous malformation and uterine artery pseudoaneurysm in literature from the past 5 years

Reports	Characteristics	US findings	Other modalities	Management	Outcomes
Lebreton *et al*. [43]	-37 years -G8P5 -4 CS and 3 curettage aspirations -AUB and increasing pelvic pain in 48 hours	CD US showed a 3 cm triangular anechoic picture in the vicinity of the CS. Color and pulsed-wave Doppler mode assessment showed a medial pulsatile arterial Doppler flow inside the scar defect, fueled by collateral of the right uterine artery, fully compatible with a diagnosis of an AVM inside. the scar defect	Not used.	-A uterine tamponade using a double-balloon catheter to stop severe bleeding. -Uterine artery embolization was performed.	-Hb from 13 g/dL to 6 g/dL. -Transfusion of 3 units of packed red blood cells. -A total hysterectomy 3 months later due to risk of recurrent massive bleeding afterwards and no envisioned pregnancy.
Li *et al*. [24]	-33 years -G3P1 -1 CS -CSP cured by UAE followed by a D&C 1 year ago -Light AUB, paroxysmal lower abdominal pain during 21 days -Failed MTX administration	-CD imaging showing enhanced vascularity surrounding and within the lesion. -Three-dimensional CD angiogram showing the AVM and the feeding and draining vessels.	T1-weighted contrast-enhanced MRI showing an enlarged vessel in the gestational mass, which is consistent with AVM.	-UAE followed by a D&C -Massive bleeding that was controlled by continuous balloon compression. -Single dose of MTX (80 mg). -Hysterectomy was discussed twice due to repeated hemorrhage; however, the patient refused. -Transvaginal removal of the ectopic gestation and repair of the uterine defect was performed.	-The fertility of the patient was preserved by transvaginal fertility-sparing surgery. -Hysterectomy was avoided.
Rambersat *et al*. [34]	-29 years -G4P2 -No CS scar -Vaginal spotting	Serpiginous vessels in the myometrium within the uterine corpus.	-MRI pelvis T1-weighted post-intravenous gadolinium administration showing serpiginous low signal flow voids in the myometrium and endometrial cavity, consistent with a uterine AVM. -Selective angiogram of the right uterine artery demonstrating filling of the uterine AVM.	Embolization.	Discharged uneventfully on day 1.

Table 2 (continued)

Reports		Characteristics	US findings	Other modalities	Management	Outcomes
Nakashololo *et al.* [17]	Case 1	-21 years -G2P0 -Severe AUB	-CD US showed hypervascular mass with bidirectional flow (Yin–Yang phenomena), color aliasing, and flow reversal. -Features of arteriovenous shunting, and a peak systolic velocity (PSV) of 71.5 cm/seconds and resistive index (RI) of 0.42.	-CTA showed a complex cystic lesion with central vivid enhancement within the posterior myometrium; multiple dilated, tortuous arterial feeders noted, from bilateral uterine and gonadal arteries. -Pre-embolization arteriogram of the bilateral uterine arteries confirmed the AVM.	Emergency endovascular intervention.	-Hb of 3.2 g/dL. -The patient was lost to follow-up.
	Case 2	-22 years -G1P0 -D & C for a miscarriage -AUB lasted in long time -Treatment with tranexamic acid and oral contraceptive pills failed	Not described.	-Extensive arteriovenous shunting was seen on pre-embolization DSA. -There were massively dilated bilateral uterine arteries with extensive branching upon entering the uterus with early venous filling. The branching arteries were nearly the same size as the main uterine artery with a corkscrew appearance.	The uterine arteries were successfully embolized with a combination of fibered and non-fibered coils (Cook), and gel foam.	Discharged uneventfully with a planned follow-up US examination after 1 month.
Glassman *et al.* [39]		-21 years -1 CS -Post-operative day 10 -Heavy AUB -Hb of 9.7 g/dL	CD US showing the "to and fro" or "yin–yang" sign, with bidirectional flow due to the swirling of blood within the pseudoaneurysm.	CT imaging of the abdomen and pelvis, with contrast confirmed the diagnosis of UAP.	-Thrombin under ultrasound guidance. -Uterine artery embolization was performed.	Discharged on post-operative day 2.
Wang *et al.* [42]		-36 years -G4P1 -1 CS -Acute massive AUB 53 days after transabdominal scar pregnancy excision	CD US showed a fuzzy boundary and abundant blood flow signal inside a 4.5 cm × 3.8 cm mass in the cervical isthmus of the right anterior wall of the uterus	-Pelvic MRI showed the mass, approximately 4.8 cm × 4.1 cm × 3.7 cm, with an abundant blood supply, in the isthmus of the cervix. -DSA images of uterine artery pseudoaneurysm.	-Selective UAE failed because of the thin and curved blood vessels. -The lesion decreased in size after transvaginal ultrasound-guided direct thrombin injection (UGTI). -Intrauterine balloon compression was used.	-Hysterectomy was carried out due to recurrent massive vaginal bleeding. -The patient recovered well after the operation and was discharged on day 141.

Table 2 (continued)

Reports	Characteristics	US findings	Other modalities	Management	Outcomes
Jha *et al.* [19]	-27 years -G4P1 -1 CS -Irregular AUB and lower abdominal pain along with persistently raised β-HCG of 37.03 UI/L following dilatation and evacuation (D&E) done for CSP of 10 weeks 4 days of gestational age	CD US revealed dilated tortuous vessels around the mass in the lower uterine segment, suggesting CSP with AVM.	-CTA revealed a mass in the lower uterine segment involving the myometrium, endometrium, and cervix with internal necrosis, and it was being fed by both the uterine arteries. -Digital subtraction angiography confirmed the diagnosis.	Bilateral uterine artery embolization achieved complete devascularization, as confirmed on post-intervention angiogram.	-Discharged the next day -Follow-up without complication.
Böckenhoff *et al.* [39]	-30 years -G1P1 -1 vaginal birth -Abdominal pain -Hb of 10.4 g/dL	CD CS revealed a double-headed anechoic structure in the center, each cavity of approximately 3–4 cm in diameter with a swirl of colors (Yin–Yang sign), and a pulse-synchronous blood flow from one cavity to the other as in arterial perfusion.	The CT scan revealed two hypodense, confluent lesions with complete contrast in the venous phase surrounded by a densely raised fringe with a total extension of about 9.4 × 6.8 cm corresponding to an intramural, double-lobed pseudoaneurysm with surrounding hematoma within the right lateral uterine wall.	An angiographic intervention was performed.	Discharged on day 10.
Butureanu *et al.* [33]	-29 years -3 CS -Heavy AUB -β-HCG of 40 mUI/mL with incomplete abortion -Hb of 6 g/dL	CD US showed a multicystic appearance and increased vascularization with different flow speeds, arterial and venous.	MRI revealed an expansive heterogeneous lesion, with numerous tubular vascular structures and indistinct borders, measuring 51 × 48 × 48 mm, suspected of being a placental trophoblastic tumor or choriocarcinoma.	-2 doses of MTX (48 and 40 mg) -A total hysterectomy, bilateral salpingectomy, and left oophorectomy, with right external iliac lymph node sampling and extended adhesiolysis.	-Persistence of the intensely vascularized area from the anterior uterine wall. -Histology confirmed retained placenta percreta and AVM.

Table 2 (continued)

Reports	Characteristics	US findings	Other modalities	Management	Outcomes
Shafwat *et al.* [35]	-36 years -G3P2 -2 CS -MTX for CSP -AUB	A bulky uterus with an ill-defined heterogeneous mass measuring 51 × 52 mm at the scar site surrounded by serpiginous tubular anechoic structures and turbulent flow on CD imaging with a low-resistance high-velocity flow pattern on spectral wave form.	-MRI showed a heterogeneous mass with central T1 hypointense signals, heterogeneous T2 signals, and no enhancement on post-contrast imaging suggesting chronic hemorrhage. -Multiple serpentine flow-related signal voids on T2-weighted images showing rapidly intense enhancement on post-contrast imaging and adjacent myometrial thinning were noted. -CT angiogram showed tortuous vascular channels within the uterus with arterial feeders. -Angiography showed supply from the branches of both uterine arteries.	Embolization angiogram.	No residual arteriovenous malformation.
Lounici *et al.* [40]	-35 years -G3P2 -1 CS -Recurrent AUB -Hb of 5.5 g/dL	Not described.	-CT showed a hyperattenuating smooth-walled sac adjacent to the left lateral wall of the cervical–isthmic region of the uterus in contact with a branch of the left uterine artery, roughly oval, measuring 07 × 09 × 11 mm, and unmodified at the venous phase, which confirmed the diagnosis of UAP.	Angiographic embolization.	Good result.
Shah *et al.* [36]	-26 years -G4P3 -Excessive AUB and lower abdominal pain -Persistently elevated beta-HCG levels -Hb from 11.2 to 7.9 mg/dL	US CD revealing signs consistent with scar ectopic pregnancy and elements of a high-flow AVM. A mosaic pattern of tangled vessels forming an AVM, characterized by a turbulent course, high velocity, and low resistance flow, consistent with AVM.	MRI pelvis shows an ill-defined T1 mixed and T2/STIR heterogeneous hyperintense and a mass lesion with multiple serpiginous T1 and T2 hypointense flow voids involving the lower uterine segment at previous CS site in anterior myometrium.	Bilateral UAE was performed.	β-hCG levels normalized after 1 week, and AUB was successfully halted. The postoperative period was uneventful.

Table 2 (continued)

Reports	Characteristics	US findings	Other modalities	Management	Outcomes
Ditchfield *et al.* [37]	-32 years -G4P1 -1 CS -Following hysteroscopy for CSEP with heavy AUB 6 months ago	A large caliber, abnormal vessel, containing both arterial and venous components with strong, high-velocity flow.	Three-dimensional reconstruction of CT angiogram demonstrating the grossly dilated uterine vein as it flows from the arteriovenous malformation.	-Embolization was performed under DSA. -Foley catheter insertion was used after −2 L of blood with hemodynamically unstable condition. Transfusion of three units of packed red blood cells, two units of fresh frozen plasma.	Hysterectomy due to recurrent heavy vaginal bleeding of 2 L after 9 days of embolization and patient's choice.
Our case	-31 years -G2P1 -1 CS -CSP treated with uterine aspiration and MTX	CD US showed the tortuous network of dilated vascular channels with turbulent flow relating to an abnormal mass in the uterine cavity.	Not used.	Surgical intervention with compressing suture and bilateral uterine artery ligation.	No complication on follow-up.

AMV, arteriovenous malformation; AUB, abnormal uterine bleeding; β-hCG, beta-human chorionic gonadotropin; CD, color Doppler; D & C, dilation and curettage; DSA, digital subtraction angiography; US, ultrasound; CTA, computed tomography angiography; CS, cesarean section; CSE, cesarean scar ectopic pregnancy; Hb, hemoglobin; MTX, methotrexate; MRI, magnetic resonance imaging; G, gravida; P, para; STIR, short TI inversion recovery; UAE, uterine artery embolization; UAP, uterine artery pseudoaneurysm

Abbreviations

AVM	Arteriovenous malformation
CSEP	Cesarean scar ectopic pregnancy
PSV	Peak systolic velocity
UAP	Uterine artery pseudoaneurysm
US	Ultrasound

Supplementary Information

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Additional file 1: Video S1. Determination of AMV.

Additional file 2: Video 2. Compressing suture using laparoscopy.

Additional file 3: Video 3. Uterine cavity examination using hysteroscopy.

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Guarantor
Phuc Nhon NGUYEN is the guarantor of the present work.

Author contributions
NDL was responsible for patient care, surgical management, supervision, and administrative procedures. PNN was involved in collecting data and contributed mainly to writing, reviewing, revising, and editing the manuscript. NDL and PNN contributed equally to this work and would like to share the first authorship. All authors read and approved the final manuscript.

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Data availability
Data sharing is not applicable to this article, as no datasets were generated or analyzed during the current study.

Declarations

Ethics approval and consent to participate
The ethical approval was naturally waived by institutional ethics committee of Tu Du Hospital and prepared in accordance with the ethical standards of the 1964 Helsinki Declaration.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The authors declare that they have no competing interests.

Author details
[1]Department of Laparoscopy, Tu Du Hospital, Ho Chi Minh City, Vietnam. [2]Department of Pregnancy Pathology, Tu Du Hospital, Ho Chi Minh City, Vietnam. [3]Clinical Research Center (CRC), Tu Du Hospital, 284 Cong Quynh, Pham Ngu Lao Ward, District 1, Ho Chi Minh City 71012, Vietnam. [4]Service de Gynécologie-Obstétrique, Maternité, Chirurgie Gynécologique, Centre Hospitalier Universitaire d'Orléans, Orléans, France. [5]Faculté de Médecine de Tours, Université de Tours, Tours, France.

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