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Arteriovenous malformation developing in a cesarean scar after termination of pregnancy – a case report and review of the literature

Arteriovenózní malformace vznikající v jizvě po císařském řezu po ukončení těhotenství – kazuistika a přehled literatury

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Summary: Uterine arteriovenous malformations (AVMs) are a rare cause of severe uterine bleeding. Uterine traumas, such as cesarean section or abortion, are known risk factors. In this article, we present a rare case of an AVM that developed in a patient with a history of a previous cesarean section and recent abortion. In this case, we review the pathogenesis, diagnosis, and treatment strategies of AVMs that develop in cesarean scars in light of the current literature. The patient presented with 2 months of vaginal bleeding that led to life-threatening anemia. Doppler ultrasound identified an AVM in the cesarean scar. Given the hemodynamic status of the patient, an emergency surgical intervention was performed and the AVM was successfully treated. This case highlights that AVM should be primarily considered in the differential diagnosis of severe bleeding in patients with a "double-hit" risk, such as a cesarean section followed by subsequent uterine trauma, and that emergency surgical intervention can be life-saving.

Key words: uterine arteriovenous malformation – cesarean scar – induced abortion – uterine hemorrhage – case report – literature review

Souhrn: Arteriovenózní malformace dělohy (AVM) jsou vzácnou příčinou závažného děložního krvácení. Traumata dělohy, jako je císařský řez nebo potrat, jsou známými rizikovými faktory. V tomto článku prezentujeme vzácný případ AVM, který se vyvinul u pacientky s anamnézou předchozího císařského řezu a nedávného potratu. V tomto případě se zabýváme patogenezí, diagnostikou a léčebnými strategiemi AVM, které se vyvíjejí v jizvách po císařském řezu, s ohledem na současnou literaturu. Pacientka se dostavila s vaginálním krvácením trvajícím 2 měsíce, které vedlo k život ohrožující anemii. Dopplerovský ultrazvuk identifikoval AVM v jizvě po císařském řezu. Vzhledem k hemodynamickému stavu pacientky byl proveden urgentní chirurgický zákrok a AVM byla úspěšně léčena. Tento případ zdůrazňuje, že AVM by měla být primárně zvažována v diferenciální diagnostice závažného krvácení u pacientek s rizikem „dvojitého zásahu", jako je císařský řez následovaný následným traumatem dělohy, a že urgentní chirurgický zákrok může zachránit život.

Klíčová slova: arteriovenózní malformace dělohy – jizva po císařském řezu – umělé přerušení těhotenství – děložní krvácení – kazuistika – literární rešerše

Introduction

Uterine arteriovenous malformations (AVMs) are abnormal capillary-free shunts between arteries and veins, which, despite their rarity, can lead to life-threatening hemorrhage [1]. In terms of etiology, acquired causes are predominant, characterized by fistulous connections that develop during the abnormal healing process after uterine trauma, such as dilatation and curettage (D&C) or cesarean section [2].

Cesarean scars, due to their fibrotic and irregularly vascularized structure compared to normal myometrium, are specific sites of trauma that create a predisposition for the development of AVM. This structural vulnerability makes scar tissue a 'hot spot' not only for AVMs, but also for other significant gynecological complications such as isthmocele and scar pregnancy [3].

The addition of secondary trauma, such as curettage, to an existing cesarean scar

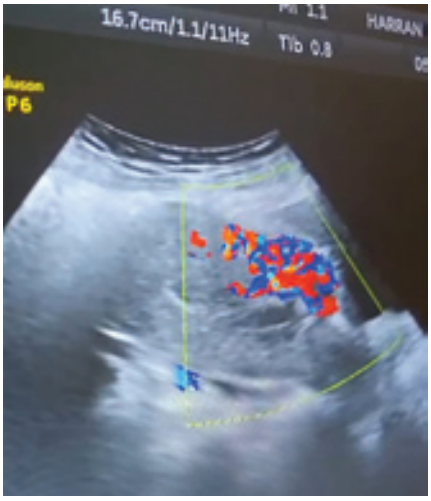

Fig. 1. Transabdominal Doppler ultrasonography image.
Obr. 1. Snímek z transabdominálního dopplerovského ultrazvuku.

creates a synergistic 'double-hit' scenario that accelerates the pathological process, rather than being simply an additive risk [4]. Indeed, the case presented here serves as a prototype for this hypothesis, with a history of a cesarean section as the 'first hit' and curettage as the 'second hit'.

The pathogenetic basis of this hypothesis lies in the hypoxic and fibrotic microenvironment created by the first trauma (cesarean section), which 'primes' the scar tissue for a pathological response [3]. The second trauma (curettage) on this 'sensitized' ground triggers uncontrolled angiogenesis, characterized by the dysregulated expression of angiogenic factors such as vascular endothelial growth factor (VEGF), instead of a normal wound healing process. The result is a pathological AVM structure in which a functional capillary network is bypassed and direct arteriovenous shunts are formed [1,5].

Although rare, these AVM cases that develop through a "double-hit" mechanism pose a potential clinical problem due to the rise in cesarean and curettage rates and carry the risk of being under-recognized [1]. Therefore, this case report aims to increase the index of suspicion for AVM, especially in at-risk patients, and to highlight diagnostic and therapeutic strategies for this rare condition.

Case report

A 33-year-old woman, gravida 2, para 1, with no known comorbidities, presented to our emergency department. Her obstetric history revealed a previous uneventful cesarean delivery. She had a history of termination of pregnancy by surgical curettage two months prior to her presentation. The patient's chief complaint was clotted and persistent vaginal bleeding, which had started after the abortion and had progressively worsened over the last month. This was accompanied by symptoms of severe anemia, such as fatigue, dizziness, and palpitations.

Upon presentation, her general condition was moderate and she was conscious. Her vital signs revealed tachycardia (pulse: 110/min) and normotension (blood pressure: 110/70 mmHg). A pelvic examination showed intense, fresh and active bleeding from the cervix. Initial laboratory investigations confirmed severe anemia with a hemoglobin level of 7.1 g/dL. Her serum beta-hCG level was negative (< 5 mIU/mL). To achieve hemodynamic stabilization, the patient immediately started a transfusion of 3 units of packed red blood cells.

Transvaginal Doppler ultrasonography identified a lesion of approximately

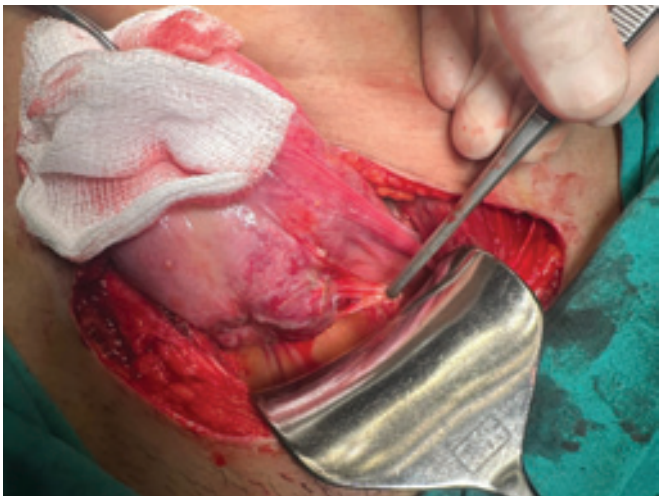

Fig. 2. Intraoperative view of the AVM of the uterine (bladder dissection).
Obr. 2. Intraoperační pohled na AVM dělohy (disekce močového měchýře).
AVM – uterine arteriovenous malformation/arteriovenózní malformace dělohy

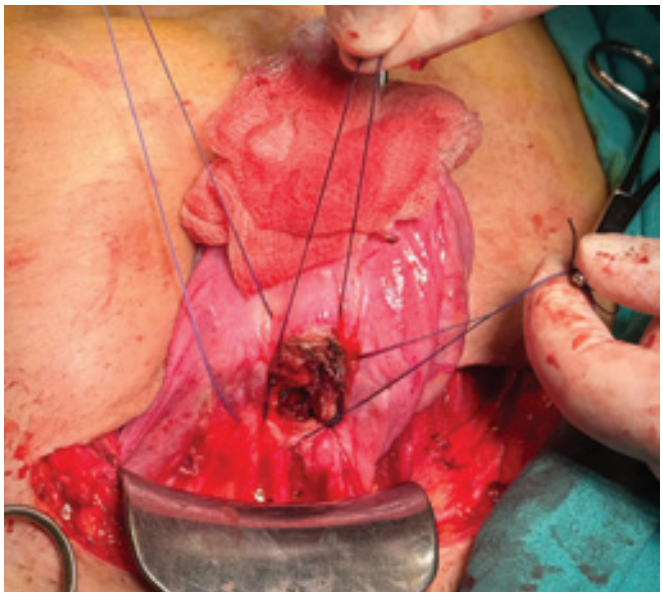

Fig. 3. Intraoperative view of the sutured defect after wedge resection.
Obr. 3. Intraoperační pohled na sešitý defekt po klínové resekci.

Tab. 1. Chronological timeline of clinical events.
Tab. 1. Chronologický sled klinických událostí.

Time point	Key clinical event/symptom	Significant findings/investigations	Interventions
unknown (past)	previous cesarean delivery	normal course	cesarean section
2 months before presentation	abortion (surgical curettage)	termination of pregnancy	curettage
1st month after abortion	onset of vaginal bleeding	intermittent, moderate bleeding	–
immediately before presentation	worsening and unremitting bleeding	heavy, clotted bleeding; fatigue	–
hospital presentation	admission to the emergency department	Hb: 7.1 g/dL, active vaginal bleeding	blood transfusion
day of presentation	Doppler ultrasonography	appearance of AVM in the cesarean scar	–
1 day after presentation	surgical intervention	excision of AVM	laparotomy with AVM resection
postoperative period	cessation of bleeding, improvement in general condition	clinical recovery	postoperative monitoring

AVM – uterine arteriovenous malformation/arteriovenózní malformace dělohy

3 × 4 cm within the myometrium of the anterior uterine wall, at the site of the previous cesarean scar, described as a "tangled mass of vessels" (Fig. 1). Color Doppler imaging showed significant hypervascularity and a mosaic pattern of colors. Spectral Doppler analysis confirmed the diagnosis of AVM with a high-velocity (Peak Systolic Velocity, PSV > 70 cm/s) and low-resistance (Resistivity Index, RI < 0.4) arterial flow pattern [6].

Due to the patient's life-threatening hemorrhage, an emergency surgical intervention was decided. Through laparotomy, the myometrial tissue containing the AVM was resected in a wedge fashion. After hemostasis was achieved, the resulting myometrial defect was repaired in two layers using delayed absorbable 2-0 polyglactin sutures (Vicryl) (Fig. 2, 3). The final hemostasis control showed no bleeding. Histopathological examination of the resected material confirmed the diagnosis of AVM. Her postoperative course was uneventful and the bleeding stopped immediately. She was discharged in stable condition on the third postoperative day, with her hemoglobin level increasing to 10.5 g/dL (Tab. 1).

Discussion

The clinical management of acquired uterine AVMs, whose pathogenetic foundations were detailed in the introduction, holds important lessons when viewed through the lens of this 'double-hit' case. In this section, the diagnostic and therapeutic aspects of our case will be analyzed by comparing them with similar cases and the current level of evidence in the literature.

Evaluation of the presented case from the "double-hit" hypothesis perspective

This case is a clinical manifestation of the "double-hit" hypothesis, the theoretical framework of which was outlined in the Introduction. The sequence of events in the patient's history strongly supports this hypothesis:

1. The previous cesarean section can be considered the 'first hit', which made the tissue susceptible to a pathological response due to the altered fibrotic microenvironment of the scar.
2. The surgical curettage applied to this sensitized ground was the 'second' hit' that triggered the AVM. Indeed, the fact that the patient's life-threatening and unremitting bleeding began exactly after this second traumatic intervention (curettage) corroborates this causal chain, which has also been emphasized in the literature [7].

Diagnostic challenges and the role of Doppler USG

In a patient with severe post-abortal bleeding, the differential diagnosis is of critical importance, as it directly influences treatment. The most commonly confused condition, retained products of conception (RPOC), is usually located within the endometrial cavity, whereas AVMs are typically found within the myometrium. In our case, the myometrial location of the lesion and the negative serum beta-hCG level aimed the diagnosis away from gestational trophoblastic diseases and RPOC, thus strengthening the suspicion of AVM [8].

At this juncture, color Doppler ultrasonography (CDUS) plays a key role as a result of its non-invasive nature and high diagnostic accuracy. Typical findings for an AVM described in the literature include a tangled mass of vessels within the myometrium showing a mosaic color pattern, and an arterial flow pattern with a high peak systolic velocity (PSV > 60–70 cm/s) and a low

Tab. 2. Principal modalities used in the treatment of uterine AVM.
Tab. 2. Hlavní metody používané při léčbě AVM dělohy.

Modality of treatment	Basic principle	Primary indications	Advantages	Disadvantages/ risks	Success rates (literature)	Effect on fertility
conservative management	observation with expectation of spontaneous regression	asymptomatic, minimal bleeding, hemodynamically stable, small AVMs	avoids interventional procedures, and is cost-effective	ongoing risk of bleeding, anxiety, and need for frequent follow-up	variable, spontaneous resolution in some cases	does not affect
medical therapy	hormonal suppression, inhibition of angiogenesis, uterotonic effect	mild-to-moderate symptomatic, hemodynamically stable patients with a desire for fertility	non-invasive, potential to preserve fertility	side effects, duration of treatment, variable efficacy, insufficient for severe bleeding	88% overall success (various agents)	generally preserved, pregnancies reported post-treatment
uterine artery mbolization (UAE)	selective occlusion of the arteries that feed the AVM	symptomatic AVMs, especially in hemodynamically stable patients with a desire for fertility, an alternative to surgery	minimally invasive, high success rate, fertility preservation potential, short hospital stay	post-embolization syndrome, groin hematoma, infection, recurrence, rarely ovarian failure, post-UAE pregnancy complications	61–91% (primary/ secondary success)	generally preserved, pregnancy rates may be similar to the general population
surgical treatment (resection/ excision)	surgical removal of the AVM (hysteroscopic, laparoscopic, by laparotomy)	UAE failure/ constraindication, places without UAE capability, life-threatening hemorrhage, selected cases	complete removal of AVM, potential for definitive cure	invasive, anesthesia risks, adhesions, bleeding, infection, risk of loss of fertility (due to uterine damage)	high, especially with complete resection	depending on uterine preservation, scar tissue formation may affect fertility
surgical treatment (hysterectomy)	complete removal of the uterus	life-threatening uncontrollable hemorrhage, failure of UAE/ resection, patient without desire for fertility, suspicion of malignancy	definitive cure, elimination of the risk of recurrence	invasive, major surgical risks, permanent loss of fertility, menopausal symptoms (if oophorectomy is performed)	100% (for AVM)	terminates fertility

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resistivity index (RI < 0.4–0.5) [6]. The PSV > 70 cm/s and the RI of < 0.4 detected in our patient were in complete agreement with these criteria of the literature, confirming This case also illustrates how a curettage performed before an AVM diagnosis can dramatically exacerbate existing bleeding; this reinforces the warning against performing a curettage before a definitive diagnosis is established [9].

Justification of the treatment strategy

The approach to uterine AVM treatment should be personalized based on factors such as the patient's hemodynamic status and desire for fertility. The literature reports successful outcomes with conservative follow-up in patients with minimal or asymptomatic bleeding and with medical treatments such as GnRH analogs or progestins in hemodynamically stable patients [10]. Currently, uterine artery embolization (UAE) is the first-line treatment for stable patients, especially those who wish to preserve fertility, with success rates exceeding 90% and no demonstrated adverse effects on subsequent pregnancy rates [7].

However, contrary to these ideal scenarios, the hemodynamic instability of our patient at presentation (tachycardia, severe anemia) and unremitting active

bleeding indicated that she was not a suitable candidate for minimally invasive methods. As noted in the literature, in the presence of life-threatening massive hemorrhage, when UAE has failed, or where facilities are limited, emergency surgical intervention comes to the forefront. Therefore, in our case, laparotomy with wedge resection to achieve immediate and definitive hemorrhage control was an evidence-based and life-saving approach [11]. For patients who have completed childbearing, in whom other treatments have failed, or in life-threatening emergencies where embolization is unavailable or unsuccessful, it is the definitive treatment, providing a 100% cure rate [12,13].

Impact of the case on clinical practice and learning points

The most important clinical takeaway from this case is the necessity of maintaining a high index of suspicion for AVM in patients with a "double-hit" risk. Since uterine AVMs are rare, treatment approaches are largely based on case series and retrospective studies rather than randomized controlled trials. This highlights the importance of multi-center patient registries for the development of evidence-based guidelines. In the future, a better understanding of the molecular mechanisms in the pathogenesis of AVM can allow for new medical therapies, while the identification of specific risk factors such as the "double-hit" could lay the groundwork for developing potential preventive strategies, such as avoiding unnecessary curettage in this group of patients (Tab. 2).

Conclusion

Uterine arteriovenous malformations that develop on the ground of a cesarean scar are rare but life-threatening causes of hemorrhage that should be considered, especially in the presence of a subsequent secondary trauma such as curettage (a "double-hit"). This case demonstrates that the possibility of AVM should be considered in at-risk patients and that color Doppler ultrasonography should be used effectively in diagnosis. Although the treatment approach must be personalized to the patient's hemodynamic status and fertility expectations, it should not be forgotten that emergency surgical intervention is a life-saving option in cases of unremitting hemorrhage. Increased awareness by clinicians of this rare condition is of critical importance in reducing morbidity and improving patient outcomes.

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