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Cesarean scar pregnancy associated with uterine artery pseudoaneurysm: a case report

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Summary

Cesarean scar pregnancy (CSP) and uterine artery pseudoaneurysms (UAPs) are associated with massive uterine hemorrhage and mortality. As a result of their low prevalence, the occurrence of CSP and a UAP in the same patient is extremely rare. The authors describe a patient who was initially misdiagnosed with trophoblastic disease by ultrasonography. The lesion had a blood-rich area of 75 × 65 × 61 mm on ultrasonography. Pelvic angiography revealed a UAP in the right side of the uterus. The patient underwent uterine artery embolization (UAE) immediately after the correct diagnosis was confirmed. Curettage was undertaken under ultrasound guidance one week postoperatively. Histopathological examination of the resected tissue revealed degenerative chorionic villi and trophoblasts with blood clots. Serum levels of beta-human chorionic gonadotropin (β-hCG) and uterine ultrasound recovered to normal levels two weeks and three months later, respectively.

Key words: Cesarean scar pregnancy (CSP); Uterine artery pseudoaneurysm (UAP); Ultrasound; Uterine artery embolization (UAE).

Introduction

Cesarean scar pregnancy (CSP) is a very rare type of ectopic pregnancy, with an estimated incidence ranging from 1:1800 to 1:2226 pregnancies [1–2]. The true incidence of CSP is not known because of the very limited literature on this subject [3], and the exact etiology and mechanism of action of CSP are poorly understood. However, it is thought that a CSP occurs if a blastocyst implants on fibrous scar tissue within a wedge-shaped myometrial defect in the anterior lower uterine segment at the site of a previous cesarean scar [4]. It is often misdiagnosed as an aborting pregnancy. CSP may result in life-threatening complications such as uterine rupture, massive hemorrhage, disseminated intravascular coagulation, and even maternal death [5]. Transvaginal ultrasonography can be used to diagnose CSP in its early stages [6] but management guidelines for this disease are lacking. Thus, each patient with CSP should be evaluated on an individual basis [5].

A pseudoaneurysm is an extraluminal collection of blood with turbulent flow that communicates with the flowing arterial blood through a defect in the arterial wall. Pseudoaneurysms typically form because of local trauma with vascular injury. If a punctured or lacerated artery does not seal completely, blood may escape and diffuse through adjacent tissues, thereby collecting in a perivascular location. If this blood maintains a communication with the patent vessel, a pseudoaneurysm may form [6, 7]. A "covert" pseudoaneurysm can easily result in massive hemorrhage, potentially resulting in the necrosis of hemorrhagic tissue and life-threatening hemorrhagic shock.

Uterine artery pseudoaneurysm (UAP) is a rare consequence of cesarean section [8, 9]. UAP can be diagnosed with angiography and color Doppler ultrasound [10, 11], which reveal a blood-filled cystic structure with a "swirling" arterial flow pattern [6, 12].

Here, the authors review a rare case of CSP associated with UAP. They also discuss the presentation, diagnosis, management strategies, and outcome for the patient.

Case Report

A 36-year-old female (G2P1) with a previous early miscarriage was admitted to the present hospital because of irregular vaginal bleeding. She had undergone a low-segment cesarean delivery 16 years previously, and had regular menstrual cycles since. Uterine curettage had been conducted 46 days previously due to an aborted pregnancy that was diagnosed by ultrasound at another hospital. Massive uterine bleeding had been noted during that procedure, which was controlled with medication.

Clinical examination revealed a normal pulse rate and normal blood pressure (110/65 mmHg). Gynecological examination demonstrated a smooth, normal-sized cervix but some dark-red blood in the vagina. The uterus was in a forward position and the uterus size was typical of a pregnancy in the third month. The uterus showed a medium mass, suitable activity, and pain upon gentle palpation. Both adnexa were normal.

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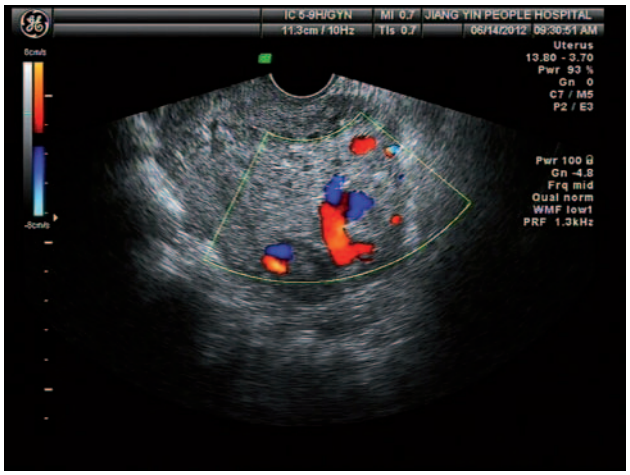

Figure 1. — Ultrasound images of a large hematoma, measuring 7.5 × 6.5 × 6.1 cm, in the anterior wall of the middle–lower uterine cavity. Color Doppler flow images (CDFI) show large serpiginous vessels within the mass.

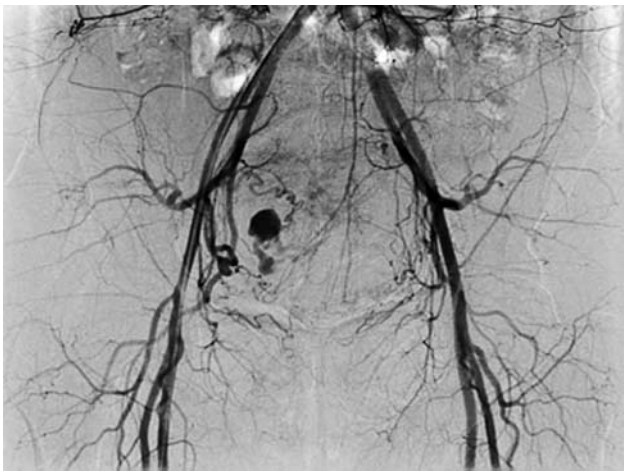

Figure 2. — Pelvic angiography reveals a uterine artery pseudoaneurysm in the right side of the uterus.

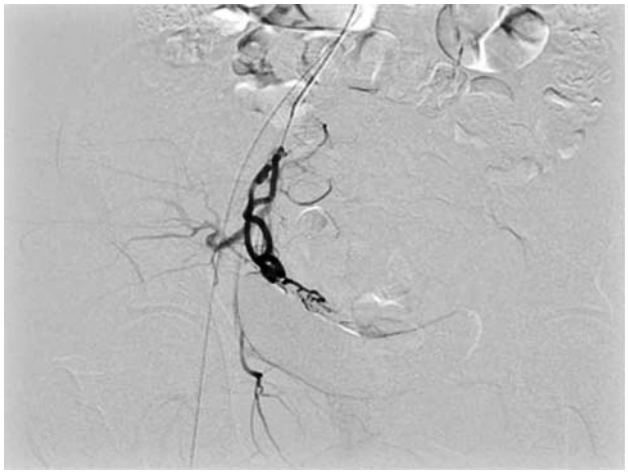

Figure 3. — UAP and extravasation of contrast agent disappeared after transmicrocatheter PVA embolization for the parent artery.

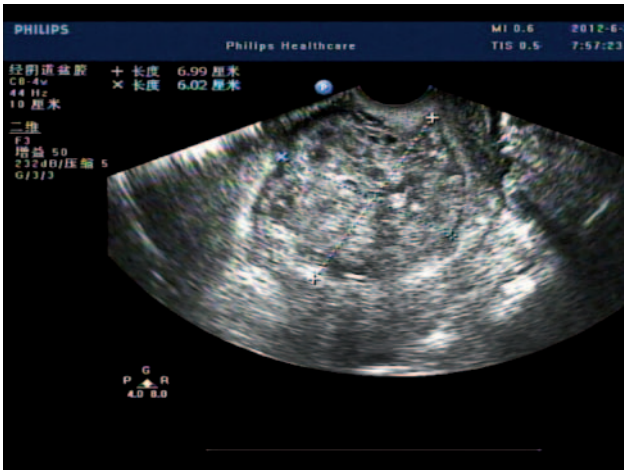

Figure 4. — Ultrasonography image one week after UAE.

Ultrasound images revealed a complex area of mixed echogenicity measuring 7.5 × 6.5 × 6.1 cm in the anterior wall of the middle–lower uterine cavity, with no clear border with the muscle layer and ≈ four mm away from the nearest placenta percreta. Color flow Doppler confirmed a large serpiginous vessel ≤ nine mm in diameter (Figure 1). The peak systolic velocity was 15.24 cm/s, and the resistance index was 0.58. According to the ultrasonographic images, a diagnosis of trophoblastic disease was considered.

After hospital admission, laboratory data revealed levels of hemoglobin (Hb) to be 85.0 g/L, beta-human chorionic gonadotropin (β-hCG) to be 1,127 IU/L, and progesterone to be 7.66 mmol/mL. A diagnosis of CSP was established according to the aforementioned findings and absence of cardiac, renal, hepatic, and blood system diseases. After appropriate counseling, the patient willing to receive uterine artery embolization (UAE) and intra-arterial methotrexate (MTX) infusion was transferred to Interventional Radiology for embolization. The next day, the patient was treated by UAE, during UAE, right UAPs were observed by angiography (Figure 2). The patient subsequently accepted embolization with an intra-arterial infusion of MTX 75 mg and polyvinyl alcohol (PVA) granules. UAP and extravasation of contrast agent disappeared after transmicrocatheter PVA embolization for the parent artery (Figure 3). The patient showed a serum level of β-hCG of 403.70 IU/L one day after UAE, which was reduced to 22.08 IU/L five days after UAE. A subsequent reduction in vaginal bleeding was observed. Repeat ultrasound imaging of the pelvis seven days after UAE showed a hematoma measuring 7.0 × 6.0 × 5.0 cm (Figure 4) without blood-flow signals within the mass. Uterine curettage was undertaken under ultrasound guidance. Blood loss during curettage was ≈ 50 mL. Histopathological examination revealed that most of the removed tissue consisted of blood clots, with inclusions of degenerative and necrotic tissue and highly degenerative chorionic villi.

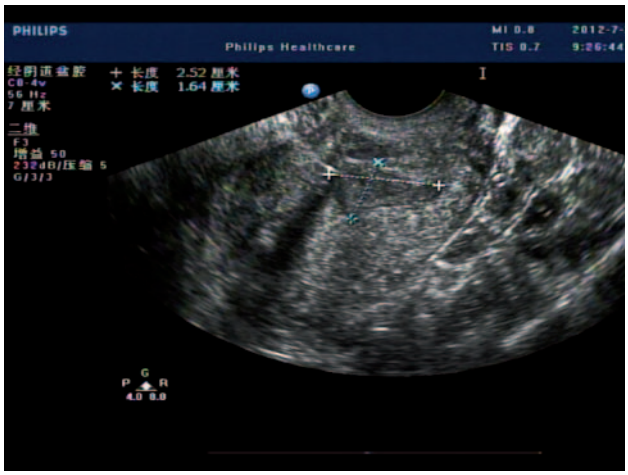

Figure 5. — Ultrasonography image one month after discharge from hospital.

Eight days after uterine curettage, the patient displayed a serum level of β-hCG of 10.4I U/L. The bleeding had stopped, and she was discharged from hospital. Weekly outpatient assessment of β-hCG levels and monthly transvaginal sonographic examinations to evaluate the size of retained products of conception were recommended [10]. Weekly outpatient clinical assessment of β-hCG until it is undetectable and monthly transvaginal sonographic examinations to evaluate the size of retained products of conception were recommended [13]. Two weeks after leaving the hospital, serum levels of β-hCG fell to normal levels. Transvaginal sonographic imaging of the pelvis one month after discharging from hospital showed a hematoma measuring 2.5 × 1.6 × 2.0 cm (Figure 5). Three months after hospitalization, ultrasound images appeared to be normal. The patient returned a normal menstrual cycle 2.5 months after hospitalization.

Discussion

In cases of CSP, the gestational sac is embedded in the myometrium, and the fibrous tissue of the cesarean scar (a long-term complication of abdominal delivery) separates from the endometrial cavity. According to recent reports, CSP may be more common than was previously thought, with an incidence of 6.1% in women with ectopic pregnancy and at least one previous cesarean delivery [2]. The lower uterine segment lacks muscle fibers, and the cesarean scar has poor levels of contractility. As a result, bleeding in a CSP can be difficult to control, and can lead to potentially life-threatening massive uterine hemorrhage or rupture.

Transvaginal color Doppler sonography is important in the early diagnosis of a CSP [1]. The criteria for CSP diagnosis was proposed by Jurkovic *et al.* [1] and Vial *et al.* [14] involved the following criteria: (i) an empty uterine cavity; (ii) an anteriorly located gestational sac at the level of the internal os, covering the visible presumed site of the previous lower uterine segment of the cesarean scar; (iii) evidence of functional trophoblastic/placental circulation on Doppler imaging; and (iv) a trophoblast between the bladder and the anterior uterine wall as a sign of deep implantation.

CSPs and UAPs can cause massive life-threatening uterine hemorrhage but are rarely seen together. To avoid massive hemorrhaging of the uterus, blind uterine curettage is not recommended once a CSP or a UAP is suspected. At the initial diagnosis by ultrasound, the clinician at the patient's previous hospital may have mistaken the CSP for an abortion due to the lower gestational sac. Blind curettage in this case could result in massive uterine hemorrhage, and iatrogenic mechanical injury could lead to the formation of a UAP. At the present hospital the patient was misdiagnosed with gestational trophoblastic disease (GTD) by ultrasonography. GTD is a type of hypersecretory hCG tumor that is secondary to vesicular moles in 60% of cases. The misdiagnosis of GTD may have been due to several reasons: a prolonged disease course, excessive bleeding, large lesion size, and an unclear boundary between the lesion and scar. Moreover, the rich internal blood supply of the lesion meant that the UAP may have been mistaken for the new vessels of GTD. The characteristic ultrasonic imaging findings for GTD are that the interior and borders of the lesion have a rich blood supply, with an abundance of high-speed, low-resistance arteries and arteriovenous fistulae [15].

Conclusion

CSPs and UAPs can cause massive, life-threatening hemorrhage of the uterus, but simultaneous occurrence of both diseases is rare. UAE is the most effective hemostatic method for the treatment of CSPs and UAPs. This method can spare the uterus and reduce the chance of death [4, 15, 16]. If a subject presents with vaginal bleeding after a uterine-cavity procedure, clinicians should be vigilant with regards to CSP and UAP. To provide timely and effective treatment, a definitive diagnosis should be made quickly via ultrasound and/or angiography.

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