

Cesarean scar pregnancy: uterine artery embolization combined with a hysterectomy at 13 weeks' gestation – a case report and review of the literature

Ciąża w bliźnie po cięciu cesarskim: Embolizacja tętnic macicznych połączona z histerektomią w 13 tygodniu ciąży – opis przypadku i przegląd piśmiennictwa

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Abstract

A cesarean scar pregnancy is a pregnancy located within the uterine muscle after previous cesarean sections. Recent years have shown a significant increase in the rate of CS and an improvement in the ultrasound diagnosis, and therefore a trend towards an increase in the rate of CSP cases has been reported in many countries.

We report on a case of CSP diagnosed using ultrasound at 5/6 weeks' gestation and confirmed by magnetic resonance imaging. The patient underwent surgical management at 13 weeks, combined with the chemoembolization of the uterine arteries. The current review aims to update the knowledge of the available treatment modalities.

Key words: **cesarean scar pregnancy / ultrasonography / magnetic resonance / embolization / treatment**

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Streszczenie

Ciąża w bliźnie po cięciu cesarskim jest ciążą zlokalizowaną w warstwie mięśniówki przedniej ściany macicy, w miejscu bliźny po cięciu cesarskim. W ostatnich latach zaobserwowano znaczący wzrost odsetka cięć cesarskich oraz udoskonalenie diagnostyki ultrasonograficznej. W wielu krajach doprowadziło to do znacznego zwiększenia liczby rozpoznawanych przypadków ciąży w bliźnie po cięciu cesarskim.

Opisywany przypadek przedstawia rozpoznanie ciąży w bliźnie po cięciu cesarskim w 5/6 tygodniu ciąży w badaniu ultrasonograficznym oraz jej potwierdzenie w rezonansie magnetycznym. W 13 tygodniu ciąży wykonano embolizację tętnic macicznych z następową histerektomią bez przydatków. Przedstawiono przegląd piśmiennictwa dotyczący aktualnych rodzajów postępowania klinicznego.

Słowa kluczowe: **ciąża w bliźnie po cięciu cesarskim / ultrasonografia /
/ rezonans magnetyczny / embolizacja / leczenie /**

Introduction

Cesarean Scar Pregnancy (CSP) is a pregnancy located within the uterine muscle, usually after one or more Cesarean Sections (CS). CSP has been described after both spontaneous and *in vitro* fertilization (IVF). [1] It is the rarest kind of ectopic pregnancy, and its incidence is estimated at 6.1%. [2] It is not correctly defined as an ectopic pregnancy because the pregnancy trophoblast is located in the lower segment of the uterine scar, in direct contact with the uterine cavity. [3]

The first CSP was reported by Larsen in 1978. [4] From 1966 to 2002 only 19 cases were reported. [5] For the next 12 years a trend towards an increasing rate of CSP cases has been reported in many countries. In 2007 there were 161 described cases, in 2010 268, and in 2011 their number increased to 751. [6, 7] Recent years have shown a significant increase in the rate of CS and an improvement in ultrasound diagnosis. In one hospital centre in China, there were 53 new cases of CSP described during the last 3 years. [8]

A previous Cesarean Deliveries (CD) increases the risk of pathological placentation. The rate of repeated CD is about 91% and the decline in vaginal deliveries after CS ranges from 9.2% to 28%. [3] CSP can occur in up to 72% of women who have had 2 or more CSs. [9] Recent studies describe the prevalence of CSP in 50% of cases after one CD, 36% after two, and 12% after three. [10]

According to the literature, the time interval between the current CSP and the previous CD can range from 6 months to 15 years. There is no consensus on how long to wait before the next pregnancy. Some authors recommend avoiding conception for 12 to 24 months. [9, 11, 12, 13]. However, there are studies where the diagnosis was made after vacuum evacuation or dilatation and curettage (D&C). [6]

The natural history of CSP may be a silent abortion. [14] Only a few of the described cases were asymptomatic in the first trimester. Less severe cases were diagnosed in the second or third trimester, usually due to a hemorrhage and uterine rupture [15, 16]

The available data suggest that the early termination of CSP is the treatment of choice. [17] The current review aims to update the knowledge of the available treatment modalities. The evidence-based guidelines for the treatment of CSP include expectant management, methotrexat therapy, local injections of methotrexat, gestational sac aspiration, dilatation and curettage,

surgical hysteroscopy/laparoscopy, laparoscopy, and uterine artery embolization. [18]

The diagnosis of CSP is a difficult task that requires experience. The primary diagnostic tool is the vaginal ultrasound. [19] The criteria for the early identification of CSP in the first trimester are as follows:

- an empty uterine cavity with clear endometrium and empty endocervical canal,
- the detection of a gestational sac within the anterior lower segment of the uterus embedded in the cesarean scar,
- an absent or thin (< 5 mm in 2/3 cases) myometrium layer between the gestational sac and bladder,
- a peritrophoblastic color Doppler flow around the sac with low-impedance (pulsatility <1), high-velocity flow (<20cm/s), a resistive index of less than 0.5 and a peak systolic/diastolic flow ratio of <3. [3, 9]
- the pathologies of the adnexa should be excluded, and there should be no detection of fluid in the Douglas pouch unless in the case of a massive hemorrhage or rupture of the uterus.

A 3-dimensional (3D) ultrasound is a useful tool in the diagnosis of CSP. The combination of multiplanar images allows the precise identification of the location, its shape and its vascularity. It also allows a differentiation from cervical pregnancy. [20] Attempts to use software for the automatic measurement of the volume of solids (VOCAL (Virtual Organ Computer-Aided Analysis)) in order to monitor changes in the vascularity of the uterus before and after successful treatment of the CSP were also described. [21]

Although transvaginal ultrasonography is the first-line method of diagnosis, magnetic resonance imaging (MRI) is complementary, especially in ambiguous cases before intervention. [6, 9, 22] MRI can be used to localize CSP, assess its volume, the possibility of penetrating the uterine wall, bladder, and abdominal cavity, and to measure the lesion volume (the fibrous tissue within the scar around the gestational sac).

In the literature, there are about 800 cases of CSP described, but still no standard therapeutic procedure has been developed. All options (surgical, pharmacological) carry a risk of serious complications. The use of arterial embolization before elective surgery to avoid bleeding is becoming more common. There are just a few descriptions of the application of this method in obstetrics.

Case report

A 37-year-old patient, gravida 4, para 4, after CS in 1996, and two vaginal deliveries, the last in 2012, was admitted in January 2013 to the Department of Obstetrics and Pathology of Pregnancy of the Medical University of Lublin due to heavy bleeding from the genital tract. The ultrasound performed with a Voluson E8, GE confirmed a single gestational sac located in the isthmus of the uterus and occupying one third of the thickness of the uterine scar after CS (0.46 mm), and a single, live fetus, with a fetal heart rate of 113 bpm and a CRL of 0.4 cm at 6 weeks 1 day (Figure 1).

The patient was informed about the life-threatening condition and advised to terminate. The patient did not consent to such a treatment, and wanted to continue the pregnancy. After stabilization of the bleeding the patient was discharged with a recommendation of treatment with progesterone and regular surveillance in the Perinathological Outpatient Clinic. At the 12th week of gestation the patient underwent non-invasive prenatal screening tests due to her maternal age. The ultrasound, performed by an obstetrician with a Certificate of Competence from The Fetal Medicine Foundation, found abnormal brain development, and an absence of parietal bones, and confirmed the previous diagnosis of the improper localization of pregnancy. The color Doppler ultrasound and VOCAL showed blood flow around the gestational sac and myometrium layer between the sac and bladder (Figures 2, 3).

The patient was admitted to the hospital at 13 weeks' gestation because of heavy vaginal bleeding. An ultrasound examination revealed a thinner uterus wall (0.24 mm) and

the possible invasion of the trophoblast into the bladder. The patient was qualified for MRI to determine the risk of in-grown trophoblasts. MRI was performed at the Department of Radiography of Medical University of Lublin, on a GE Optima 360 1.5 T in the sequences SSFSE + FATSAT, FIESTA, frFs, GRE, FGR, a T2-weighted image, T2*- and T1-dependent, frontal, sagittal and cross-sections. The examination confirmed the overgrowth of the muscle of the uterus by the trophoblast, and found trophoblast adherence to the bladder wall which did not exclude the possibility of it growing in. (Figure 4). Surgical management was proposed to the patient, and she consented to a hysterectomy only after a prior uterine artery embolization (UAE). The procedure was performed at the Department of Radiography under a local anesthetic given in the right femoral artery by the Seldinger method. The initial angiography showed the presence of extensive branches of uterine arteries and unaffected branches of internal iliac arteries (Figure 5). Both uterine arteries were catheterized selectively and closed using Embozene. The catheter was then withdrawn and an internal iliac artery embolization was performed using corks with a gelatin sponge ('torpedoes'). The final control angiography showed no flow of the described branches. Immediately after embolization, the patient underwent a hysterectomy without adnexa. Intraoperatively, after the separation of the peritoneum, we found that the uterine scar had ruptured and that the amniotic sac was protruding through the scar after CS into the abdominal cavity (Figure 6). There were no complications during the treatment. The histopathology report (No. 88465) confirmed the diagnosis of CSP with evidence of

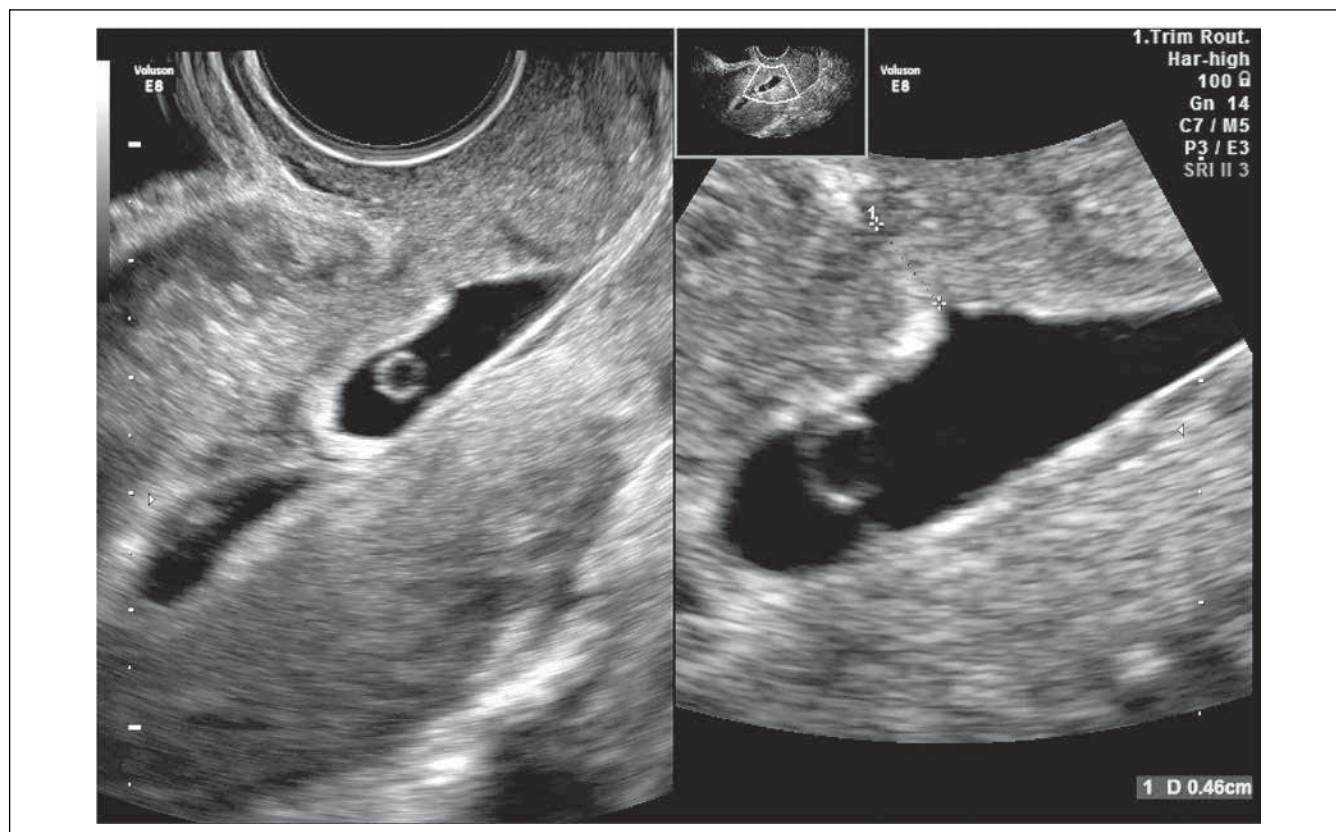


Figure 1. Transvaginal ultrasound image of a cesarean scar pregnancy at 6 weeks. A gestational sac is visible within the myometrium with the endometrial and cervical canals both empty. The thickness of the uterine wall is 0.46 cm.

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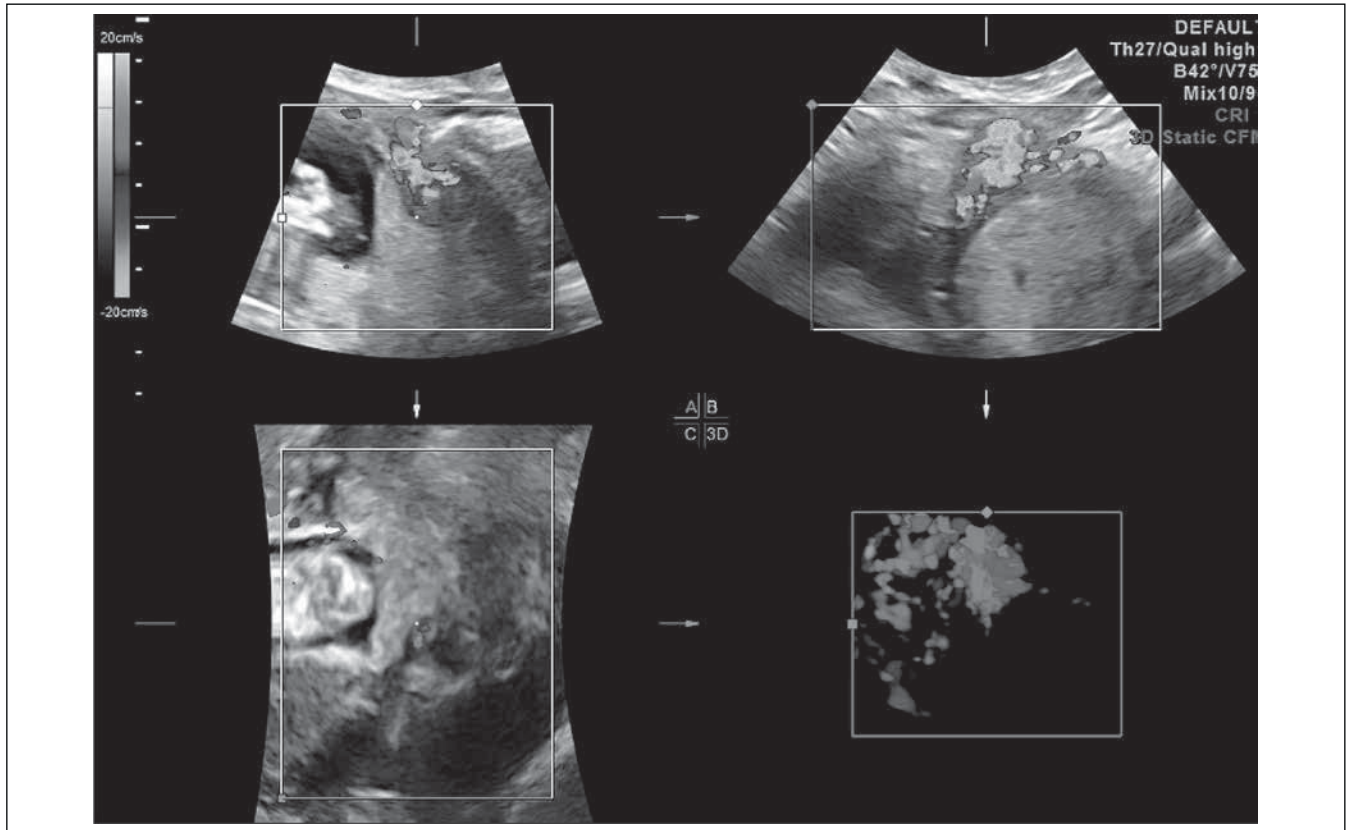


Figure 2. Evaluation of the vascular supply of the cesarean scar pregnancy at 13 weeks' gestation with a 3D transvaginal ultrasound with VOCAL software.

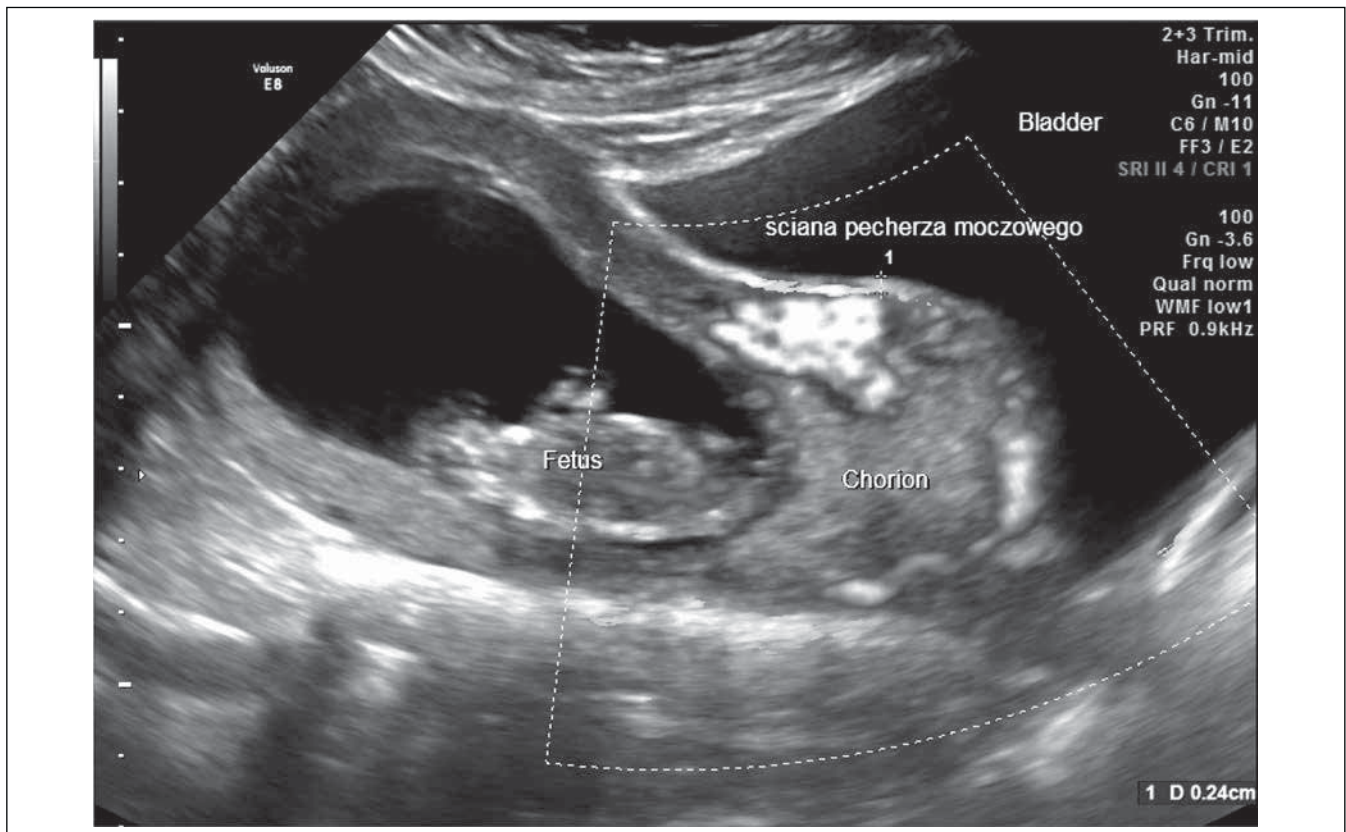


Figure 3. An ultrasound performed at 13 weeks' gestation, using an abdominal probe. The apparent vascularization confirms the invasion of the trophoblast into the cesarean section scar and towards the bladder. The myometrium thickness is 0.24 cm.

cyto- and syncytiotrophoblast cells within the muscle of the uterus. The fetus in the macroscopic examination had no parietal bones. The family did not agree to a fetal autopsy. The patient's postoperative period was uneventful and she was discharged from hospital on the seventh day.

Discussion

We describe a case of CSP which is a serious type of ectopic pregnancy. The diagnosis was made at 5/6 weeks' gestation with ultrasound. According to the literature, the mean-time for the recognition of CPS take place between the 33rd and 94th gestational day. [6, 9, 18, 22, 23]. However, a proper diagnosis remains a challenge for many doctors. In the study by Zhang et al., all 17 described cases were incorrectly diagnosed as normal intrauterine pregnancies, abnormal intrauterine pregnancies such as inevitable abortion, missed abortion or trophoblastic disease, or cervical ectopic pregnancies. [22] Among the 100 cases of CSP presented by Yu et al., 7 were misdiagnosed, with one diagnosed as choriocarcinoma. [24] Several studies point out that the diagnosis of CSP was made after massive bleeding occurred after performing curettage. [24] The ability to diagnose CSP early is needed in order to successfully administer treatment.

In our case, the CSP was diagnosed by a transvaginal color Doppler ultrasonography which met all the criteria raised by Vial et al. [19] The uterus cavity and cervical canal were empty, with the implantation of the gestational sac in the scar tissue, and with a growth towards the bladder. The progression of the trophoblast tissue towards the bladder wall had been shown by a reduction in the thickness of the uterine wall from 4.6 mm at 6 weeks' gestation to 2.4 mm at 13 weeks'.

The diagnostic process was extended with a 3D transvaginal ultrasound and VOCAL. The angiographic rendering showed vascularization around the gestational sac, between the uterus wall and the bladder. [3] In other studies, this proved to be a reliable method of differential diagnosis. To confirm the diagnosis, a computer tomography angiography is invaluable. [25]

The initial diagnosis, in our case, was confirmed by MRI. In other studies, the sagittal, coronal and transverse section of T1- and T2-weighted magnetic resonance imaging sequences were used to show the gestational sac embedded in the anterior lower uterine segment, and to assess the possibility of a myometrial invasion in the bladder. [6, 9, 18]

A verified diagnosis should lead to the termination of the pregnancy. A delay in treatment can result in uterine rupture, or the hemorrhaging of the placenta percreta. In some cases, the patients decided to continue with their pregnancies. The earliest gestation at which CSP has been diagnosed was at 5 weeks, and it was managed conservatively. [16] At 37 weeks' gestation, due to a massive hemorrhage, a caesarian section with a subtotal hysterectomy was performed, and the final histopathological diagnosis was anterior placenta previa/accreta. Other researchers describe conservative management from 6-7 until 35-37 weeks' gestation, usually resulting in an urgent cesarean hysterectomy with or without bladder repair due to hemorrhaging. [15, 26]

The Doppler flow showed an invasion of vessels into the cesarean scar but the pregnancy endured until 36 weeks. After elective CS and an uncontrolled hemorrhage a total hysterectomy was performed. Many cases of spontaneous uterine rupture have been reported in the first and second trimester. [27]

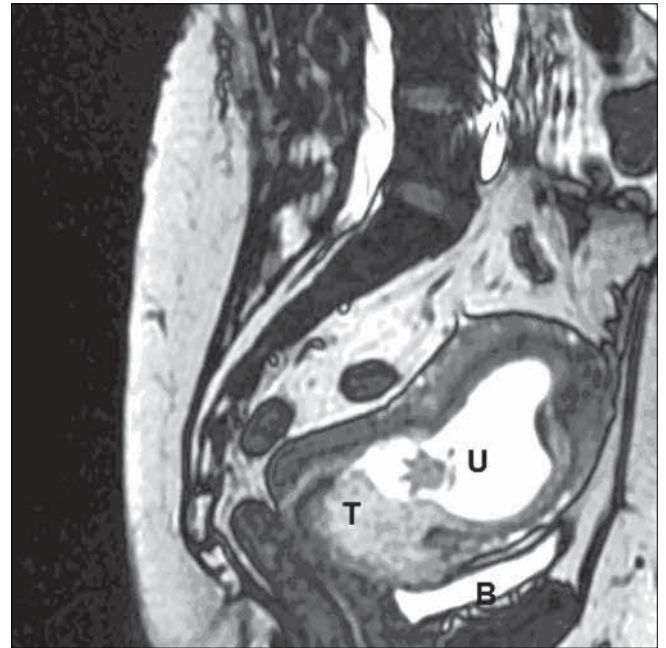


Figure 4. A magnetic resonance image of the cesarean scar pregnancy at 13 weeks, showing the infiltration of the trophoblast into the uterine wall and towards the bladder. U – uterus, T – trophoblast, B – bladder.



Figure 5. An angiogram of the left uterine artery with the extravasation of the contrast before uterine artery embolization.

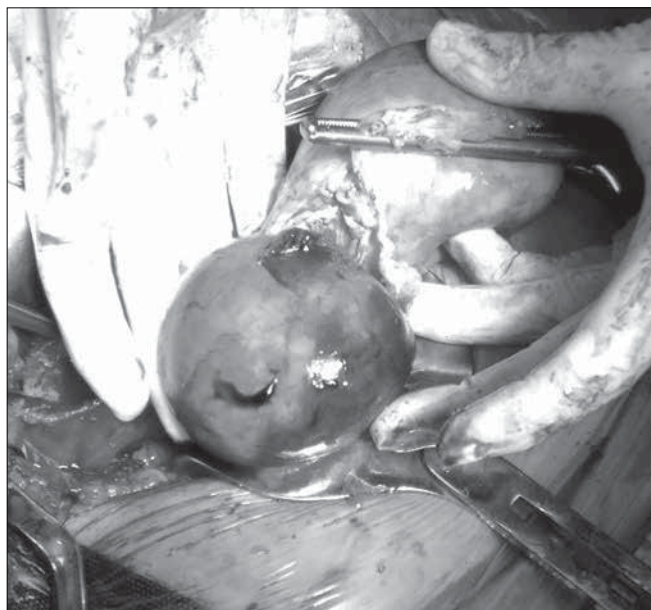


Figure 6. Ruptured uterine scar with an amniotic sac protruding into the abdominal cavity.

The decision regarding treatment should be made by the patient and the clinician based on expertise and experience. In our case, the patient did not agree to terminate the pregnancy at 6 weeks. At 13 weeks' gestation, when the abnormalities of the fetus were discovered, vaginal bleeding occurred and, with the risk of an in-growing trophoblast in the bladder, she consented to surgical treatment. Before the 7th week abortion should be considered, but afterwards methotrexat and/or surgical options should be preferred. [28] Other researchers measured the proceeding of the myometrial thickness between the gestational mass and bladder. [12] Their experience of 53 patients suggests that the intrauterine approach (hysteroscopy) is safe in cases with a myometrial thickness ≥ 2 mm, and a gestational mass < 3 cm in size, and a gestational sac protruding towards the uterine cavity. The UAE is a method of treating CSP alone, or in combination with the administration of methotrexat, before or after uterine curettage, a hysterectomy, or as life-saving management during massive bleeding. Based on a 15-patient retrospective study, the conclusion on the effectiveness of uterine artery embolization was confirmed. [23] Eleven patients were offered preventive UAE combined with methotrexat before curettage. The other four were treated with emergency UAE for uncontrollable hemorrhage after curettage. The study by Zhang et al. investigated, among other things, the total blood loss after D&C preceded by or without UAE. [29] The result showed significantly greater bleeding during D&C before bilateral uterine artery chemoembolization.

There is no reliable data on the role of the interval between the previous CD and the risk of CSP. In our case, the patient had CS 17 years before admission to hospital, and during that period had 2 uncomplicated vaginal deliveries. In the literature, the time period between CS and the actual diagnosis of CSP ranges from 11 to 159 months. [12, 13, 17, 18, 23] However, there are no studies presenting a patient with physiological deliveries during this period of time.

Conclusion

Blind dilatation and curettage should not be recommended for CSP.

All pregnant women with a previous history of uterine scars (CS, myomectomy) should undergo a transvaginal ultrasonography examination to determine the localization of the gestational sac at the early stage of pregnancy.

A multicentre collaboration and evidence-based studies are needed for creating recommendations for the treatment of SCP.

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