

CASE REPORT

A rare case report of heterotopic cesarean scar pregnancy in the 8th week of gestation that was managed successfully by exploratory laparotomy with dilation and curettage

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Key Clinical Message

Swift and precise identification of heterotopic cesarean scar pregnancy, coupled with standardized treatment approaches for handling possible serious complications, form an essential component in reaching favorable outcomes for patients experiencing this rare type of pregnancy.

Abstract

Heterotopic pregnancy (HP) denotes a form of multiple gestation where intrauterine and ectopic pregnancies coexist. Cesarean scar ectopic pregnancy, on the other hand, involves the implantation of a fetus over the previous cesarean scar. This condition poses a significant risk of uterine rupture, which may lead to serious health complications, and even death. We report a case of a fit 37-year-old woman with two previous cesarean deliveries who was diagnosed with a heterotopic cesarean scar pregnancy at 8 weeks gestation following symptoms of lower abdominal pain and delayed menstruation. Both pregnancies demonstrated cardiac activity and the portion of the myometrium located between the bladder wall and the gestational sac was noted to exhibit considerable thinness. The patient underwent an exploratory laparotomy coupled with dilation and curettage and recovered uneventfully. The proper management of a HCSP requires timely diagnosis through ultrasonography. Early diagnosis allows for immediate intervention to prevent complications such as uterine rupture or potentially lethal bleeding.

KEYWORDS

dilation and curettage, heterotopic cesarean scar pregnancy, laparotomy, ultrasonography

1 | INTRODUCTION

Heterotopic pregnancy (HP) is characterized by the existence of intrauterine and ectopic pregnancies occurring simultaneously.¹ This is a rare yet serious condition that can be spontaneous or resultant from assisted reproductive technology (ART).² While spontaneous HP has a reported frequency of 1 in 50,000–1 in 10,000,^{3,4} ART-related cases of HP have been estimated to occur in 0.2%–1% of patients.⁵ Heterotopic cesarean scar pregnancy (HCSP) involves the occurrence of a cesarean scar pregnancy (CSP) accompanied by intrauterine pregnancy (IUP), which poses a high risk of catastrophic complications such as uterine rupture and massive hemorrhage.^{6–8} An extremely low incidence of HCSP has been reported during spontaneous cycles. Nevertheless, due to the rising occurrence of cesarean section delivery and the expanding recourse to ARTs, the prevalence of HCSP is anticipated to increase.^{9–11} Due to the considerable risk for fetal and maternal morbidity and mortality, timely, and precise diagnosis of HCSP is vital.¹² Medical imaging technology, a baseline serum human chorionic gonadotropin (HCG) concentration, and the history of a cesarean section form the basis of the clinical diagnosis.¹³ Patients' reproductive needs, hemorrhage risk, and imaging classification determine the therapeutic strategy. To diagnose, the most common imaging methods are traditional 2D and 3D color/power Doppler ultrasound, contrast-enhanced ultrasound, and magnetic resonance imaging (MRI).¹⁴ As the first-line imaging method for CSP, ultrasound is considered the preferred diagnostic method. Ultrasound can detect CSP with a sensitivity of 84.6%.¹⁵ When transvaginal ultrasound (TVS) is performed, the probe is close to the cervix, preventing intestinal gas interference, and displays the GS's location without requiring bladder filling. By performing this procedure, one can find out the size, and shape of the gestational sac, the thickness of the myometrium around the scar, and embryo, as well as the yolk sac and the heartbeat in the gestational sac. Additionally, transabdominal ultrasound (TAS) can demonstrate the relationship between the gestational sac and the bladder when properly filled.¹⁶ In color and power Doppler ultrasounds, abundant low-impedance flow signals can be obtained to determine the blood flow to the mass.¹⁰ In terms of diagnosing CSP, determining treatment options, and determining prognoses, TVS, TAS, and Doppler ultrasound appear to be of significant value together.¹⁷

In the management of HCSP, preserving the coexistent IUP presents a significant challenge. Guidelines for managing HCSP while preserving the IUP are not universally standardized due to the rarity of this condition.¹⁸

The typical method is to terminate the implantation located within the scarred area, if deemed necessary, at the potential expense of terminating the IUP.⁷ The available literature reveals a range of techniques for managing this condition, with medical, and surgical approaches being two distinct options. The surgical interventions, which may involve laparoscopic or hysteroscopic excision of the masses, have been linked with potential complications like the loss of pregnancy and preterm delivery. In our research, we detail a case of HCSP, which was addressed through a combination of exploratory laparotomy and dilation and curettage (D&C) procedures.

2 | CASE DESCRIPTION

A healthy 37-year-old woman, with a history of two previous cesarean sections due to breech presentation and repeated cesarean delivery 12 and 5 years earlier, without any significant complication, presented to our facility with sudden lower abdominal pain and delay in menstruation. The patient was hemodynamically stable and the abdominal examination was not positive for tenderness, guarding, or rebound tenderness. No blood was detected in the vaginal examination and the cervix was closed. The woman's lab results, including a beta-human chorionic gonadotropin (β -hCG) measurement of 174,025 milli-international units/mL, were all within the accepted range. Transabdominal and Transvaginal ultrasonography revealed a dichorionic diamniotic twin gestation, with cardiac activity, and crown-rump length appropriate for both fetuses for 7 weeks and 5 days of gestation. There was no apparent difference in size between the embryos (the crown-rump length of the fetuses was 1.38 cm). Almost the same sac sizes were observed for heterotopic and intrauterine pregnancies, the gestational sac of the IUP, which was identified in the endometrial cavity was 26 mm in diameter and the gestational sac of the HP which was found in the anterior uterine isthmus, specifically at the site of the earlier cesarean scar was 28 mm in diameter. The thickness of the myometrium at the implantation site of the heterotopic sac was measured at 2.5 mm (Figures 1 and 2), and rich blood flow (in color Doppler ultrasound examination) amidst the gestational sac and the wall of the bladder (Figure 3).

3 | METHODS

The above findings were suggestive of a HCSP. The patient and her husband received guidance regarding the management options and potential maternal and fetal complications associated with the continuation of the

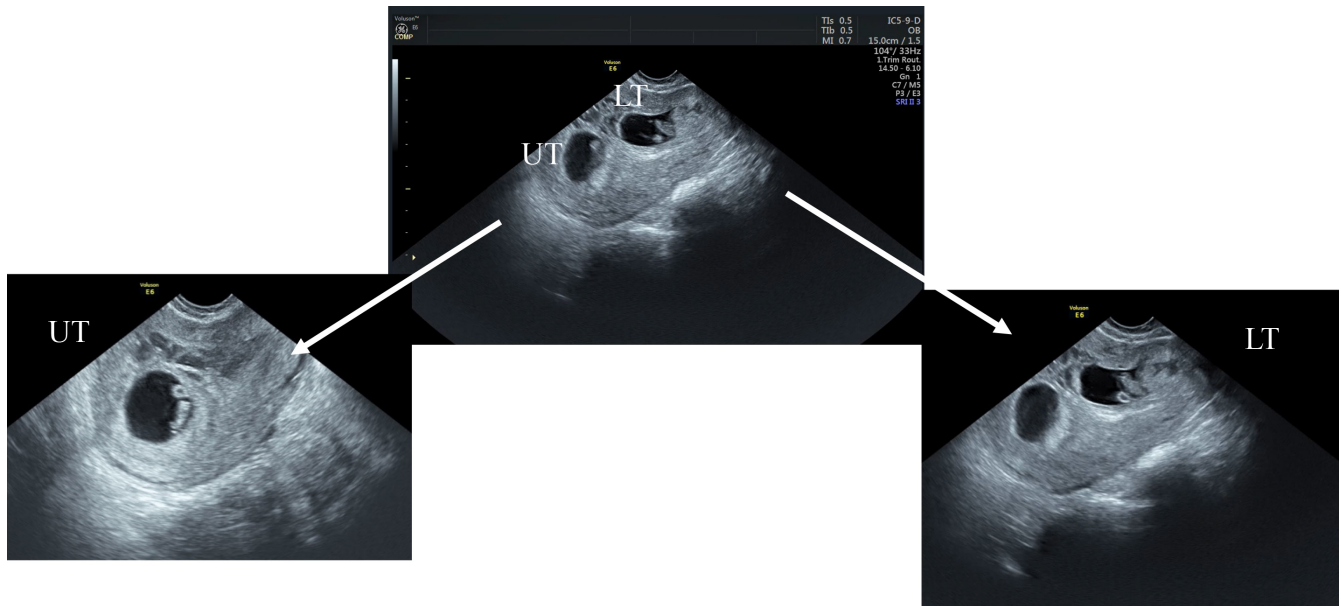
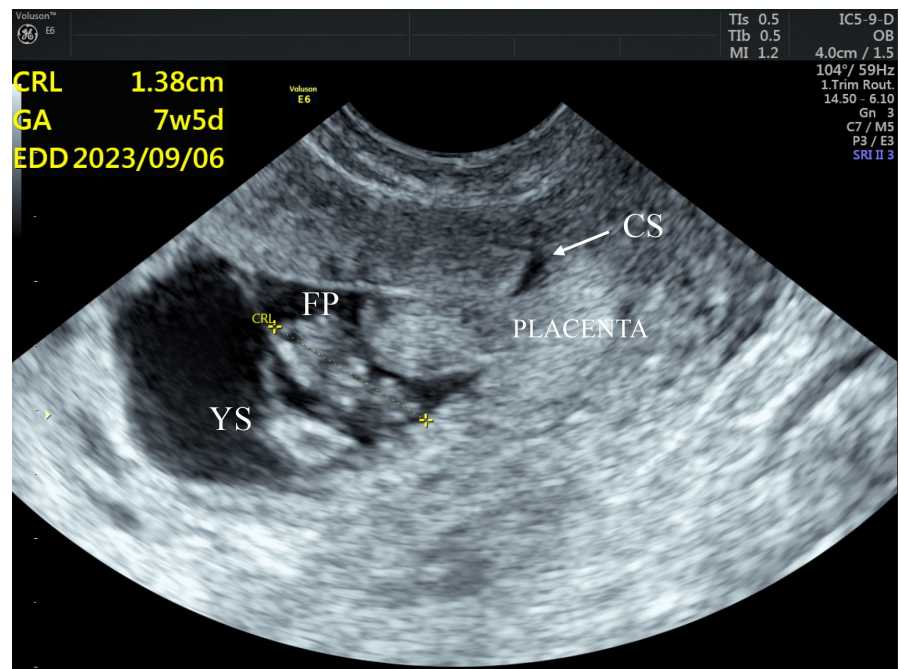


FIGURE 1 Dichorionic diamniotic pregnancy, with one sac implanted at the cesarean scar. Transvaginal ultrasonography; sagittal view showing two separate intrauterine gestational sacs with two yolk sacs and alive embryos; the upper twin (UT): normally implanted in the endometrial cavity; the lower twin (LT): abnormally implanted at the site of the previous cesarean section scar.

FIGURE 2 Magnified TVS sagittal view shows the ectopic gestational sac at the lower uterine segment at the site of the cesarean scar (CS) with a yolk sac (YS) and a fetal pole (FP) with cardiac activity and a crown-rump length (CRL) of 1.38 cm with estimated gestational age (GA) of 7 weeks and 5 days. The ectopic gestational sac extends into the cesarean scar (CS), occupying more than one-half thickness of the lower uterine segment. The overlying myometrium is thinned out (between cursors).



pregnancy, such as excessive bleeding, abortion, and the need for subsequent hysterectomy. After thorough counseling, the couple did not want to preserve the intrauterine gestation. The obstetrician arranged for the patient to undergo an exploratory laparotomy coupled with a D & C procedure. An exploratory laparotomy was performed to observe the Isthmus uteri and if necessary, undergo a partial, or total hysterectomy. However, the curettage was uneventful, no perforations were caused, and no incisions were made. The bleeding during surgery was about 500 mL. US was performed the following day which

demonstrated complete evacuation of the uterus and β -hCG was dropped to 68,344 milli-international units/mL 48 h after the operation.

4 | CONCLUSION AND RESULTS

The patient was discharged on postoperative Day 3 and her next follow-ups were unremarkable. The proper management of a HCSP requires timely diagnosis through ultrasonography. Early diagnosis allows for immediate

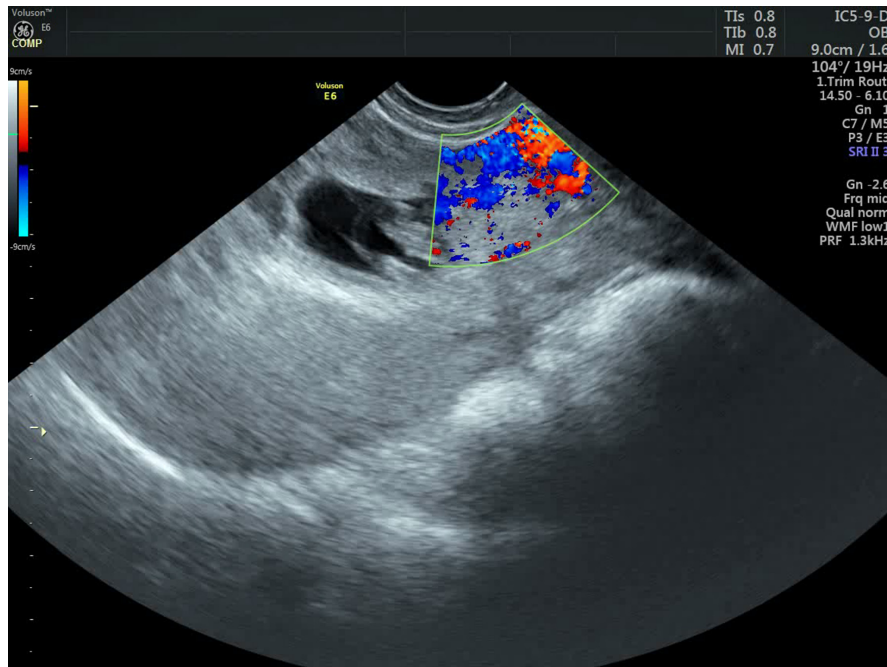


FIGURE 3 Transvaginal color Doppler ultrasonography shows rich vascularity at the implantation site of the lower twin within the previous cesarean scar.

intervention to prevent complications such as uterine rupture or potentially lethal bleeding.

5 | DISCUSSION

HCSP is recognized as one of the least common forms of heterotopic pregnancies, requiring careful observation of a potential IUP.^{8,19} It has been documented that approximately 1 in 30,000 deliveries conventionally encompasses HP. However, with the advent and continued use of ART, there has been an observable increase in the occurrence of HP, which is currently approximated at 1%.^{20,21} Several theories have been put forward in an attempt to elucidate the origin of this condition. The most plausible hypothesis posits that the blastocyst makes its way through the uterine wall through a small, non-continuous pathway. This could potentially be a consequence of damage incurred during a cesarean section, other forms of uterine surgery, or following manual placenta removal. Even without a history of uterine surgery, in vitro fertilization could occasionally lead to this outcome. CSP may demonstrate a symptom-free clinical trajectory or present with specific clinical signs like unusual vaginal bleeding and/or abdominal discomfort, or sudden abdominal pain due to uterine rupture.¹ Given the potentially fatal complications, such as severe bleeding and rupture, it is vital to diagnose, and manage this condition early. In the early phases of pregnancy, to initially detect a CSP, the primary imaging method recommended is ultrasound scan.²² Typically, sonographic imaging can identify an increase in the size of

the cesarean scar in the lower segment, as well as either a disparate mass or a distinct gestational sac linked to it. There are situations where a vulnerable myometrium, positioned between the bladder wall and the gestational scar, can be seen before rupture.²³ The main sonographic features indicative of a scar pregnancy diagnosis include (i) A vacant uterus, (ii) an unoccupied cervical canal, and (iii) the positioning of the gestational sac at the foremost region of the isthmic portion of the uterus, accompanied by a slender layer of myometrium located between the bladder and the sac.^{24–27} Moreover, a break in the front wall of the uterus can be detected via a sagittal view of the uterus. This is achieved when the ultrasound beam's direction crosses through the amniotic sac.²⁸ Doppler flow is also important for determining implantation location.² Doppler flow evaluations can differentiate a viable pregnancy located in the scar region from a non-viable intrauterine pregnancy. This distinction affects treatment strategies. If an intrauterine pregnancy proves non-viable, the gestational sac appears devoid of vasculature, indicating its separation from the implantation site. Conversely, if a CSP maintains viability, the gestational sac displays a well-vascularized appearance in Doppler examinations. The gestational mass within the scar region exhibits a low-impedance flow rate (pulsatility index <1) and a high speed (peak velocity >20 cm/s).²⁹ Various investigators have also noted that the blood flow resistance index is below 0.5, and the peak value ratio of systolic-to-diastolic flow is less than 3.²³ TVS, when used in conjunction with color Doppler analysis, exhibits 85% diagnostic sensitivity for CSP detection.³⁰ Recently, some clinicians have begun utilizing

three-dimensional (3D) ultrasonography and 3D Power Doppler imaging.³¹ Based on their findings, employing multiplanar views with 3D-rendered images can enhance diagnostic precision in such circumstances. In cases where diagnosis becomes intricate or challenging, Magnetic Resonance Imaging may also prove advantageous.³² HCSP handling is usually complex, especially if the woman wants to preserve her current intrauterine embryo.³³ The treatment protocol specific to HP involving CSP is not universally recognized and established. Various strategies are available for managing HCSP, including watchful waiting, medical intervention, and surgical termination. Successful cases of expectant management have been documented in the medical literature. However, given the unfavorable prognosis for an uncomplicated full-term pregnancy, this approach is generally not advised.¹² There are several approaches to CSP, including conservative approaches (feticide using potassium chloride (KCl) or hyperosmolar glucose, methotrexate (MTX) locally or systemically, and embryo aspiration), surgical approaches (dilatation and curettage, hysteroscopy, laparoscopy, or laparotomy), or combination approaches.³³ In numerous case studies, MTX is an effective treatment for ectopic pregnancies. However, there are concerns that its use alongside an intrauterine pregnancy might cause teratogenic effects.⁶ Because of methotrexate's potential teratogenicity and embryotoxicity, its use is not recommended in HCSP. There are a few reports of HCSP with viable pregnancies treated with KCl injections, which are traditionally used for fetal reduction in multiple pregnancies. It is important to note, however, that KCl is associated with certain inherent risks, including abdominal discomfort, miscarriage, excessive vaginal bleeding, preterm birth, a need for further surgery, and spontaneous rupture of the amniotic membranes, which can result in chorioamnionitis.⁶ Moreover, embryo aspiration may only be successful in the early stages of pregnancy when the embryo has not yet developed.³⁴ The foremost operative treatment option for ectopic CSP usually entails the explicit extraction of the ectopic mass situated at the site of the earlier cesarean scar.³⁵ Some scholars have suggested a surgical approach as the primary management strategy for an HCSP.¹⁸ In a few cases, hysteroscopic removal of the CSP was found to be successful and safe as an alternative treatment for preserving viable intrauterine gestation.³⁶ Methotrexate and hysteroscopic resection have also been reported to manage term pregnancy following CSP.³⁷ It has also been reported that hysteroscopic termination of a heterotopic cervical ectopic pregnancy can preserve a concurrent intrauterine pregnancy.³⁸ It has been reported that laparoscopic

removal of heterotopic CSP ectopic mass can lead to good pregnancy outcomes.³⁹ Nevertheless, laparoscopy should be performed simultaneously with anatomic dissection, trimming of unhealthy tissues, and repair of the uterine defect following the principles of laparotomy.⁴⁰ Open laparotomies are popular because of their superior surgical control and the expanded operational field they offer. As a result, excessive bleeding can be effectively controlled more effectively with an open laparotomy.³⁵ HIFU is a recently developed technique in which acoustic waves are converted to thermal energy when temperature reaches 65°C.⁴¹ Studies have investigated HIFU treatment for CSP in the past few years, an approach that is considered to be effective and noninvasive.⁴²⁻⁴⁴ A further benefit of HIFU is that it can reduce the risk of intraoperative bleeding.⁴¹ In general, HIFU has been successfully applied to studies without complications. As a treatment option for CSP, endovascular surgery, and uterine artery embolization are also proven to be effective.^{45,46} The patient presented to our facility with mild symptoms, and an ultrasound examination identified heterotopic CSP with one viable intrauterine embryo aged 7 weeks and 5 days. A detailed explanation of HCSP's potential hazards and complexities, as well as management strategies, was given to the couple. Since the couple did not intend to maintain the normally implanted viable intrauterine pregnancy, as well as the thin myometrium between the gestational sac and the bladder wall posing a significant risk of heavy bleeding, it was decided to perform an exploratory laparotomy with D & C. It was a successful surgery, the uterus was preserved without severe blood loss, and the patient recovered smoothly.

AUTHOR CONTRIBUTIONS

Mahsa Karbasi: Conceptualization; data curation; project administration; supervision; writing – original draft. **Reza Aletaha:** Conceptualization; investigation. **Ramin Ahangar-Sirous:** Conceptualization; investigation. **Amir Honarmand Alamdari:** Conceptualization; investigation. **Esmail Gharepapagh:** Supervision; writing – review and editing. **Sahar Rezaei:** Conceptualization; supervision; validation; visualization; writing – review and editing.

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DATA AVAILABILITY STATEMENT

All available data are presented in this article and if more explanations are needed, contact the corresponding author.

ETHICS STATEMENT

Not applicable.

CONSENT

The patient provided his written informed consent to publish and allowed for the publication of any accompanying images. Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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