Case Report

Successful Full-Term Delivery via Selective Ectopic Embryo Reduction Accompanied by Uterine Cerclage in a Heterotopic Cesarean Scar Pregnancy: A Case Report and Literature Review

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Abstract: Heterotopic cesarean scar pregnancy (HCSP) is a combination of cesarean scar pregnancy (CSP) and intrauterine pregnancy (IUP). Cesarean scar pregnancy is accompanied by life-threatening complications, such as uterine rupture and massive bleeding. Herein, we present a case of HCSP treated with selective potassium chloride injection into the CSP under ultrasonography in association with uterine cerclage to control vaginal bleeding; this led to a successful IUP preservation and full-term delivery. Additionally, we will review several previous reports on HCSP management, including our case.

Keywords: heterotopic cesarean section scar pregnancy; ectopic pregnancy; selective embryo reduction; arteriovenous malformation

1. Introduction

Heterotopic cesarean scar pregnancy (HCSP) is a rare cesarean scar pregnancy (CSP) combined with an intrauterine pregnancy (IUP), and is accompanied by life-threatening complications, such as uterine rupture and massive bleeding [1,2]. The incidence of HCSP in natural pregnancy is extremely low; however, the number of cesarean sections and the expansion of assisted reproductive technology (ART) have gradually increased [3]. Nevertheless, the preservation of concurrent IUP and fertility remains a challenge because of the absence of a standard protocol for HCSP management [2,3]. Herein, we present a case of HCSP that was treated to preserve the IUP and the patient’s fertility. In the first trimester, a selective ectopic embryo reduction in the CSP was performed using intrathoracic potassium chloride (KCl) injection and embryo aspiration. In the second trimester, the remaining gestational tissue growth in the CSP, as well as the occurrence of vaginal bleeding, was controlled via a uterine cervical cerclage. Finally, full-term delivery was successfully achieved without uterine arterial embolization or hysterectomy by repeated cesarean sections.

2. Case Report

A 36-year-old woman (gravida 2, para 1) was transferred from a local hospital because of a cesarean ectopic pregnancy with IUP. The patient underwent in vitro fertilization-embryo transfer (IVF-ET) using two ova. Two years ago, she delivered a baby via a lower segment cesarean section. Ultrasonography at 6 + 1 gestational weeks (GW) revealed two gestational sacs; one in the uterine fundus and the other in the anterior uterine isthmus (Figure 1a,b). Both had fetal cardiac activity, and the mother had no vaginal bleeding or abdominal pain.
The patient decided to undergo selective embryo CSP reduction to preserve the normal fetus. Under spinal anesthesia, a uterine sound was inserted to reach the CSP (the anterior uterine isthmus) in the uterus at 6+3 GW under ultrasonographic guidance (Figure 2a). A 20 cm long 18-gauge spinal needle, which was bent at an equal angle to the sound, was then guided along to reach the CSP (Figure 2b). About 0.1 mL of 2 mEq/mL KCl was slowly loaded via the bent 18-gauge spinal needle into the fetal heart of the CSP (Figure 2c). After cardiac arrest of the CSP, the expired embryo was completely aspirated without affecting the placenta around the CSP. No immediate complications were observed in either the mother or the normal IUP after treatment.

The following day, sonography showed an absence of cardiac activity in the CSP, whereas the IUP was alive. Five days after the procedure, a 4 × 1.7 cm placenta with hematoma was detected at the reduction site using sonography, while the normal fetus in the uterine fundus was stable (Figure 3).

A sonographic examination at 10 GW revealed that the placenta around the reduction site had grown into the uterine cervix, resulting in intermittent vaginal spotting and a shortening of the cervical length. Considering a cervical length of less than 10 mm and the protrusion of the remnant placenta into the internal os (Figure 4a), a uterine cervical cerclage was inserted at 12 GW by placing the cervical intruding placenta inside the uterine cavity. The purpose of the cerclage was to control the growth of the remaining placenta in the CSP, as well as vaginal bleeding (Figure 4b,c). McDonald operation with double ligations using braided polyester thread (Ethibond™, Ethicon, New Jersey) were implemented in
the cervical cerclage [4]. A previous retrospective study reported that the braided thread suture in the cervical cerclage showed an improvement in neonatal survival, the prevention of preterm birth before 28 GW, less PPROM, and maternal febrile morbidity, compared to Mersilene tape (Mersilene™, Ethicon, Somerville, NJ, USA). [5]. Ultrasonography at 24+3 GW showed dilated and tortuous blood vessels encompassing the lesion (10 × 6 × 3 cm³ in volume), suggestive of an enlarged arteriovenous malformation (AVM) (Figure 4d). During the antenatal period, there were no severe complications, including preterm labor and short cervical length.

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Figure 3. Transvaginal ultrasonography examination at 7+1 GW. *—intrauterine gestational sac; †—RGT; CSP—cesarean scar pregnancy; GW—gestational weeks; RGT—remnant gestational tissue.

Figure 4. Management of arterio-venous malformation after selective CSP embryo reduction. (a) Ultrasonography to visualize RGT with cervical shortening at 10³ GW before cervical cerclage. The RGT went into the uterine cervical internal os. (b) Transvaginal ultrasonography at 12³ GW after the cerclage. (c) Effect of cervical cerclage role on HCSP management (details are written in the discussion section). (d) Ultrasound examination at 24+3 GW. Enlarged arterio-venous circulation was observed in the demised CSP (†); asterisk (*) indicates the fetal foot. CSP—cesarean scar pregnancy; GW—gestational weeks; RGT—remnant gestational tissue.
The delivery was performed at another hospital for private reasons. A healthy female baby weighing 2415 g was delivered via elective cesarean section at 37+6 GW. After the delivery of the baby, massive bleeding developed at the site of the CSP. The RGT was removed, and the bleeding focus was controlled by multiple sutures. Her vital signs were stable with a red blood cell transfusion. She did not need further intensive care. The RGT was histologically confirmed as an AVM.

3. Discussion

Heterotopic cesarean scar pregnancy is one of the rarest heterotopic pregnancies, and it requires the careful management of a viable IUP [3,6,7]. The incidence of heterotopic pregnancy is estimated to be 1 in 30,000 deliveries. However, ART has increased the incidence of heterotopic pregnancies to 1% [3,8]. Attempts have been made to identify the cause of CSP at a molecular level. For example, a previous study showed that the endometrial expression of the integrin β3 subunit and leukemia inhibitory factor (LIF) was positively correlated with endometrial receptivity and embryo implantation [9]. In particular, their expression in the cesarean scar was significantly higher than in the endometrium of the uterine cavity. Heterotopic cesarean scar pregnancy poses a higher risk of antenatal events, such as vaginal bleeding, fetal demise of the IUP, and uterine rupture [1,7,10–12].

Our comprehensive literature survey of 46 HCSP cases published in journals confirmed that 32 patients reported HCSP related to ART (Table 1) [1–3,6,7,10,12–31]. Advances in ultrasonography have facilitated the early detection of HCSP, even during the embryonic period [6,22]. Although a standard protocol for managing HSCP remains inconclusive, most procedures focus on the selective reduction of the ectopic CSP [2,31]. The treatment modality for selective embryo/fetal reduction usually involves either an ultrasound (US)-guided intervention or surgical intervention (or both). Among the 42 cases, from which four artificial abortions were excluded, 19 (45%) cases were expectant management and 16 (38%) cases were US-guided interventions (Table 1). Surgical intervention accounted for only seven cases (17%). The US-guided interventions included the injection of embroyocidal drugs, gestational sac (GS) aspiration, or a combined procedure. The US-guided interventions were performed between 8\textsuperscript{+2} and 10\textsuperscript{+2} GW (median: 8\textsuperscript{+4} GW). Potassium chloride was used in all injections, except for one case where methotrexate (MTX), which can cause teratogenicity to the normal IUP, was co-administered [18]. The KCI injection facilitated the spontaneous regression/detachment of the demised CSP; however, 12 cases among the US-guided interventions reported that the remnant gestational tissue (RGT) still existed. The persistence of RGT in CSP can lead to various complications, such as vaginal bleeding, probably due to its vascular characteristics [1,2,7,13–15,18,20–22,25]. Indeed, seven cases reported some problematic concerns (Table 1). For example, four cases documented vaginal bleeding after the intervention. Three cases described the development of RGT into AVM, among which one case was accompanied by a morbidly adherent placenta (MAP) that eventually required a forced hysterectomy [25]. The other RGT-to-AVM case involved a friable vascular mass with dilated vessels in a repeat cesarean section, in which uterine artery embolization was performed to control bleeding [22].
Table 1. Literature review of heterotopic cesarean scar pregnancy.

| Reference | Conception/Previous CS (n) | Diagnosis Modality/GW/Symptoms or Event | Cardiac Activity of CSP/IUP | Management/GW | RGT | Antenatal Event | Pregnancy Outcome |
|-----------|---------------------------|----------------------------------------|-----------------------------|---------------|-----|-----------------|-------------------|
| Salomon [13], 2003 | ART/1 | TVUS/8/None | Yes/Yes | US-guided intervention (KCL injection)/NM | Persistent | PROM | CS at 36 GW, live female, 2800 g, RGT excision during CS |
| Yazicioglu [14], 2004 | Spontaneous/1 | TVUS/6/2 /VB | Yes/Yes | US-guided intervention (KCL injection)/7-2 | Spontaneously disappeared | PROM | CS at 30 GW, live male, 1530 g, RGT detachment without complications |
| Hsieh [10], 2004 | ART (twin IUPs + CSP)/2 | TVUS/6/VB | Yes/Yes | US-guided intervention (EA)/NM | Spontaneously disappeared | Preterm labor | CS at 32 GW |
| Wang [1], 2007 | ART/3 | TVUS/7/None | Yes/Yes | US-guided intervention (KCL injection)/NM | Persistent | Preterm labor | CS at 35 GW, live male, 1820 g, bilateral internal iliac artery ligation due uterine bleeding, RGT excision during CS |
| Demirel [12], 2009 | N/M/1 | TVUS/6/5 /VB | Yes/Yes | Surgical intervention (laparoscopy)/NM | Removed | None | CS at 38 GW, live singleton |
| Taskin [15], 2009 | N/M/1 | TVUS/8/4 /VB | Yes/Yes | US-guided intervention (KCL injection)/9 | Persistent | Preterm labor | CS at 34 GW, live female, 2310 g, RGT excision during CS |
| Wang [16], 2010 | ART/1 | TVUS/7/VB | Yes/Yes | Surgical intervention (hysterotomy)/7 | Removed | None | CS at 39 GW, live male, 3250g |
| Gupta [17], 2010 | ART/4 | TVUS/6/1/None | Yes/Yes | US-guided intervention (EA)/6-3 | Persistent | None | Termination at 12 GW due to trisomy 13 |
| Litwicka [18], 2011 | ART/1 | TVUS/6/None | Yes/Yes | US-guided intervention (KCI + MTX injection)/8 MTX + leucovorin (used for abortion)/NM | Persistent | Placental abruption | CS at 36 GW, 1990 g male, Miller syndrome |
| Dueñas-Garcia and Young [19], 2011 | Spontaneous/3 | TVUS, MRI/5/None | Yes/Yes | US-guided intervention (KCI injection + EA)/NM | None | None | CS at 38 GW, live singleton, subtotal hysterectomy due to postpartum bleeding |
| Ugurlucan [3], 2012 | ART/1 | TVUS/6/VB | Yes/Yes | US-guided intervention (KCI injection + EA)/NM | None | None | CSP miscarriage at 8-4 GW, VB and protruding RGT |
| Bai [20], 2012 | ART/1 | TVUS/7/6 /VB | Yes/Yes | Expectant | Persistent | CSP miscarriage at 8-4 GW, VB and protruding RGT | CS at 36-4 GW due to preterm labor, live male, 2950 g |
| Uysal [21], 2013 | Spontaneous/2 | TVUS/8/None | Yes/Yes | US-guided intervention (KCL injection)/NM | Persistent | Preterm labor | CS at 35 GW, live female, 2480 g, incomplete uterine rupture, RGT excision during CS |
| Lui [22], 2014 | ART/1 | TVUS/5/VB | Yes/Yes | US-guided intervention (repeated EA)/NM | Persistent | None | CS at 37 GW, live female, 2660 g, RGT with AVM, selective UAE |
| Kim [6], 2014 | Spontaneous/2 | TVUS/5/5/None | Yes/Yes | Expectant | Persistent | None | CS at 37-5 GW, live twins, bladder adhesion, placenta accreta, bilateral uterine artery ligation |
| Armbrust [23], 2015 | ART/2 | TVUS/7/None | Yes/Yes | Surgical intervention (laparotomy)/NM | None | None | CS at 37-5 GW, live singleton, 2895 g |
| Reference          | Conception/Previous CS (n) | Diagnosis Modality/GW/Symptoms or Event | Cardiac Activity of CSP/IUP | Management/GW                                      | RGT                       | Antenatal Event | Pregnancy Outcome                                                                 |
|--------------------|----------------------------|-----------------------------------------|-----------------------------|---------------------------------------------------|---------------------------|-----------------|----------------------------------------------------------------------------------|
| Yu [2], 2016       | ART/1                      | TVUS/11/None                            | Yes/Yes                     | US-guided intervention (KCl) /16+4                | Persistent               | PPT, accreta     | CS at 37+6 GW, live male, 2890 g, profuse vascularization with bladder adhesion, RGT excision during CS |
| Czuczwar [24], 2016| NM/1                       | TVUS/6/None                              | Yes/Yes                     | US-guided intervention (KCl injection) /7         | None                     | None            | CS at 37 GW, live male, 3060 g                                                  |
| Linenberg [7], 2016| NM/3                       | TVUS/10\(^2\)/AP, intraperitoneal hemorrhage | Yes/Yes                     | Surgical intervention (laparoscopy, laparotomy) /10\(^2\) | Persistent               | Uterine rupture | CS at 23+1 GW, live female, 423 g                                                |
| Vetter [26], 2016  | Spontaneous/1              | TVUS/5/VB                                | Yes/Yes (too early)         | Surgical intervention (laparotomy)/NM             | None                     | None            | CS at 37+1 GW, live female, 3479 g                                               |
| Miyague [25], 2018 | NM/1                       | TVUS, MRI/6/None                         | Yes/Yes                     | US-guided intervention (combined KCL injection + EA)/NM | Growth with vascularity  | RGT growth and AVM and MAP formation | Hysterectomy                                               |
| Vikhareva [27], 2018| NM/1                       | TVUS/13/None                             | None/Yes                    | US-guided intervention (KCl injection) /10+6      | Disappeared at 18 GW     | None            | VD at 39 GW, live male, 2985 g                                                  |
| Tymon-Rosario [28], 2018 | NM/2               | TVUS/NM/None                             | Yes/Yes                     | US-guided intervention (KCl injection) /10+6      | N/M                      | Septic shock    | Hysterectomy after UAE, D&C                                                     |
| Ashwini J Authreya [31], 2021 | ART/1 | TVUS/7\(^+\)/6/None                        | Yes/Yes                     | Hyperosmolar glucose injection and EA/8+2, transcervical D&C | Disappeared at 20 GW     | None            | CS at 38 GW, a term healthy baby                                                 |
| Zheng-Yun Chen [29], 2021 | Spontaneous/1                | TVUS/8/None                               | Yes/Yes                     | Hyperosmolar glucose injection and EA/8+2, transcervical D&C | Abortion (D&C)          | Vaginal bleeding | CS at 34+2 GW, a healthy baby PROM D&C + UAE                                   |
| Ouyang [30], 2021  | ART/1                      | TVUS/6/VB                                | Yes/Yes                     | US-guided intervention (KCl injection) /NM        | Persistent               | Vaginal bleeding | CS at 14+2 GW, a healthy baby PROM D&C + UAE                                   |
| Ouyang [30], 2021  | ART/1                      | TVUS/6/2/VB                              | Yes/Yes                     | Hyperosmolar glucose injection and EA/8+2, transcervical D&C | Persistent               | Persistent      | Hysterectomy excision of the CSP due to placenta accreta at 8 GW Hysteroscopic removal of RGT |
| Ouyang [30], 2021  | ART/1                      | TVUS/6/VB                                | Yes/None                    | HIFU /7                                           | Persistent               | None            | Miscarriage of IUP at 7 GW                                                      |
| Ouyang [30], 2021  | ART/1                      | TVUS/5/VB                                | Yes/Yes                     | D&C/13                                            | Disappeared at 13 GW     | CSP miscarriage at 13 GW                                                        |
| Ouyang [30], 2021  | ART/1                      | TVUS/6/VB                                | Yes/Yes                     | Exemptant                                         | Disappeared at 20 GW     | CSP miscarriage at 13 GW                                                        |
| Ouyang [30], 2021  | ART/1                      | TVUS/6/VB                                | Yes/Yes                     | Exemptant                                         | NM                      | CSP miscarriage at 13 GW                                                        |
| Ouyang [30], 2021  | ART/1                      | TVUS/5/6/VB                              | Yes/Yes                     | Exemptant                                         | NM                      | IUP miscarriage at 20 GW                                                        |

**Table 1. Cont.**
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| Reference          | Conception/Previous CS (n) | Diagnosis Modality/GW/Symptoms or Event | Cardiac Activity of CSP/IUP | Management/GW | RGT      | Antenatal Event                  | Pregnancy Outcome                  |
|--------------------|---------------------------|----------------------------------------|------------------------------|---------------|---------|---------------------------------|-----------------------------------|
| Ouyang [30], 2021 | ART/1                     | TVUS/6/VB                              | Yes/Yes                      | Expectant     | NM      | IIIOC                           | Induced abortion at 22 GW          |
|                    |                           |                                        |                              |               | Persistent | CSP miscarriage at 10 GW       | CS at 37 GW, live male, 2600 g    |
| Ouyang [30], 2021 | ART/2                     | TVUS/6/None                            | Yes/Yes                      | Expectant     | Disappeared at 22 GW            | PROM                             | CS at 39 GW, live female, 2900 g  |
|                    |                           |                                        |                              |               | Persistent | CS at 39 GW, live female 3900 g | CS at 24 GW                        |
| Ouyang [30], 2021 | ART/1                     | TVUS/6/None                            | None/Yes                     | Expectant     | Persistent | CS at 39 GW, live singleton, 2600 g |
| Ouyang [30], 2021 | ART/1                     | TVUS/6/None                            | None/Yes                     | Expectant     | Persistent | CS at 39 GW, live male, 3150 g  |
|                    |                           |                                        |                              |               | Persistent |                      | Removed                           |
| Laing-Aiken [32], 2020 | Spontaneous/1             | TVUS/9/VB                              | Yes/Yes                      | Surgical intervention (D&C, laparotomy)/9 | Removed | PPROM                           | CS at 28 GW, live male, 1200 g, bilateral uterine artery ligation |

AP—abdominal pain; ART—assisted reproduction techniques; AVM—arteriovenous malformation; CS—cesarean section; CSP—cesarean section pregnancy; D&C—dilation and curettage; EA—embryo aspiration; GW—gestational weeks; IIIOC—incompetent internal os of cervix; HIFU—high-intensity focused ultrasound; IUP—intrauterine pregnancy; KCL—potassium chloride; MAP—morbidly adherent placenta; MRI—magnetic resonance imaging; NM—not mentioned; PROM—premature rupture of membrane; PPROM—preterm premature rupture of membrane; TVUS—transvaginal ultrasonography; UAE—uterine artery embolization; VB—vaginal bleeding.
The fundamental surgical intervention for ectopic CSP is the direct excision of the ectopic mass at the cesarean scar via an open laparotomy, hysteroscopy, laparoscopy, or dilatation and curettage. Our literature review identified seven cases of surgical intervention. The surgical removal of CSP is a feasible way to prevent antenatal complications, such as vaginal bleeding and RGT growth [10,30]. Moreover, pelviscopic excision can reinforce the lower uterine segment [30]. An open laparotomy would provide more security to the previous scar because the extent of the operation field becomes broader and the operator can handle profuse bleeding more easily [21]. It is of note that the surgical approach for RGT removal should be considered cautiously in terms of preserving fertility. One patient experienced uterine rupture, and the normal fetus ended with early preterm birth before 24 weeks, even after surgical repair [5]. Another case involved laparoscopic excision after a US-guided intervention, owing to the growth of an ectopic mass and MAP-like sonographic findings. However, the surgery resulted in a hysterectomy because of uncontrolled bleeding [23].

Another option for HCSP was expectant management (19 cases) [4,18,25,28]. Nine cases confirmed the absence of a heartbeat in their CSPs, whereas seven cases reported live births and two cases did not. Among the two deaths, one was an induced abortion due to the premature preterm rupture of the membrane in early IUP, and the other was a uterine rupture accompanied by severe bleeding at 12 GW. The latter case involved a laparotomy for repair, but the IUP did not survive [28]. Among the 10 cases with a heartbeat in their CSPs, three reported miscarriages in the CSP but live IUPs were delivered [28]. Additionally, one case documented massive bleeding at eight GW during expectant management and underwent hysteroscopic excision [28]. Two vital babies from CSPs were successfully delivered in two cases at 37 and 40 GW [28]. However, severe postpartum bleeding due to placenta accreta occurred in one case, which was managed by the excision of the uterine anterior lower segment and uterine artery ligation. The other patient also had focal placenta accreta [28]. One case reported the implementation of expectant management for a successful delivery of twins [4]. Nevertheless, a recent study on the association between poor obstetric outcomes and HCSP demonstrated that the gestational age at treatment and a higher number of previous CSs were related to antepartum and postpartum hemorrhage, irrespective of the treatment mode [29].

4. Conclusions

As described before, US-guided intervention can lead to RGT persistence, which can develop into AVM, accompanied by various complications, including vaginal bleeding, weakness of the scar site, or incomplete scar rupture. Sonography-guided selective CSP embryo reduction, which was successfully employed in our case, has been recommended for HCSP management because of its easy manipulation, high IUP success rate, and fewer complications, although it cannot exclude RGT persistence [30]. Herein, we implemented a cervical cerclage after a selective CSP embryo reduction to prevent cervical shortening caused by AVM; this eventually controlled vaginal bleeding and promoted a successful full-term delivery. It can be speculated that surgical cerclage may tighten the loosened cervical canal, counteract the outward pressure formed by AVM, and preclude massive hemorrhage during the pregnancy (Figure 2c). Although a standard procedure has not been established, the first application of cervical cerclage in HCSP management in our case was helpful in managing fertility and IUP survival, as well as in reducing complications.

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