Iran Red Crescent Med J. 2017 September; 19(9):e60000. doi: 10.5812/ircmj.60000.

Published online 2017 August 22. **Case Report**

Heterotopic Cesarean Scar Pregnancy in a Non-assisted Fertility: A Case Report

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Received 2017 February 10; **Revised** 2017 April 07; **Accepted** 2017 June 03.

Abstract

Introduction: Cesarean rate increased in recent decades worldwide. One of the consequences of the increased cesarean rate and repeat cesarean is the significant increase in cesarean scar pregnancies (CSPs). Diagnosis of a CSP is more difficult when there is a heterotopic pregnancy in a non-assisted pregnancy.
Case Presentation: The patient was a 34-year-old G5P2L2Ab2 referred for spotting in Shahidan Mobini hospital, Sabzevar, Iran in 2016. She had a history of 2 cesareans and 2 abortions. Three ultrasounds were performed showing a gestational sac in the lower segment of the uterus with different diagnoses: 1) with hemorrhage over it, 2) with the 2nd gestational sac over it, which was diagnosed as missed abortion, and 3) with an echo-free and irregularly region supporting the 2nd sac or a clot in the lower part of the uterus. The increased local vascularity suggested a level of placenta accreta, partial mole, or trophoblastic reaction. Since the first diagnosis was missed abortion, curettage was performed. Due to the continuation of severe bleeding, abdominal hysterectomy was performed. The patient was discharged in good condition after 3 days.
Conclusions: Heterotopic CSP does not have any specific symptoms, which caused it to be easily misdiagnosed. Physicians should use precise diagnostic tests in case of controversial test results.

Keywords: Cesarean Section, Repeat, Ectopic Pregnancy, Twin Pregnancy, Uterine Rupture, Hysterectomy

1. Introduction

Cesarean rate increased in recent decades worldwide (1). One of the consequences of the increased cesarean rate and elective repeat cesarean is the significant increase in cesarean scar pregnancies (CSPs). It is speculated that cesarean scar pregnancy results from the incomplete healing of the lower uterine segment in the previous cesarean, which leads to invasion of the new trophoblasts of subsequent pregnancy (2). A review of 39 cases of CSP showed that it may also be the consequence of continued growth of residual pregnancy mass after incomplete abortion or curettage to manage CSP with gestation sac (3). CSP can be a life-threatening condition, which may result in uterine rupture and uncontrollable hemorrhage (4). Heterotopic CSP (HSCP) is a serious form of ectopic pregnancy. It is possible that the embryo in the uterus has the potential to develop, while the other develops in the cesarean incision (4) and threaten the life of the mother and normally implanted embryo. Moreover, more than half of the HCSPs are asymptomatic (5), making its diagnosis more difficult. HSCP is rare. The current study found only 18 case-reports till 2017. Result of a systematic review identified 14 patients with HSCP in 2014. It included 6 spontaneous pregnancies and 8 in vitro fertilization-embryo transfer (5). Therefore, it is more probable that it follows assisted reproductive technologies (ART) in females with a previous cesarean (6). In the present case, HCSP followed a non-assisted pregnancy.

2. Case Presentation

The patient was a 34-year-old healthy female and resident of Chesham village with an unclear last menstrual period (LMP), 2 previous cesareans (1999 and 2007) and 2 abortions (2011 and 2016), which resulted in curettage (Table 1). According to 3 ultrasound examinations, she was hospitalized with missed abortion diagnosis and the possibility of placenta accretes in the lower segment of the uterus in Shahidan Mobini hospital, affiliated to Sabzevar University of Medical Sciences, Sabzevar, Iran on 26 Nov, 2016. Shahidan Mobini hospital is a specialized state referral hospital where childbirth, obstetrics, and gynecologic conditions are treated and pregnant and postpartum mothers receive specialized care. Overall, there are 7 wards and 110 beds in this hospital.

Table 1. Patient's Obstetrics History and Ultrasound Findings

Variables	Test Result
Age, y	34
Gravida	5
Parity	2
Abortion	2
Mode of 2 previous deliveries	Cesarean
The interval from the last caesarean to HCSP, y	10
The interval from the last curettage to HCSP, mo	10
First ultrasound result	A gestational sac (8 weeks) without a fetal pole in the cervical canal with a sub chorionic hemorrhage (40 × 15 mm) over it
Second ultrasound result	A gestational sac (7 weeks + 6 days) with a fetal pole and the 2nd gestational sac (34 mm) over the first sac in the uterine cavity without a fetal pole
Third ultrasound result	A gestational sac (7 - 8 weeks) with irregular margin and no fetal pole and an echogenic debris (7 - 8 weeks) suggesting the 2nd sac without a fetal pole or a clot
Hospitalization days	4

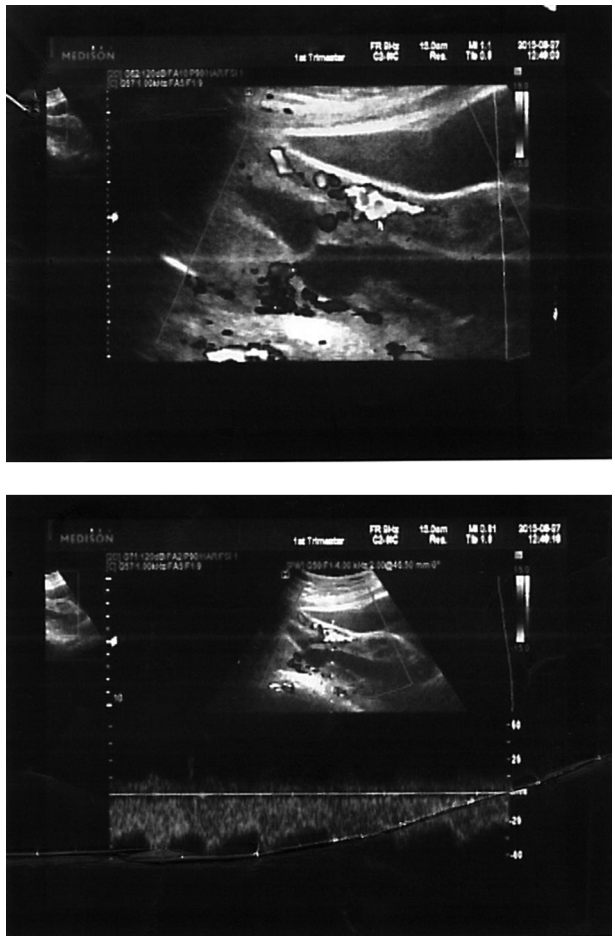

Figure 1. Transabdominal Doppler Shows an Increased Local Vascularity in the Superior Portion of the Uterine Body Lower Part (the 3rd Ultrasound)

Results of the first ultrasound on 03 Nov, 2016 indicated a gestational sac with 28 mm diameter (GA = 8weeks) and irregular margin without a fetal pole in cervical canal lower than a normal site with a sub chorionic hemorrhage (40 × 15 mm) over it. The 2nd ultrasound was performed on 14 Nov 2016 in which the gestational sac (54 mm, 7 weeks + 6 days) with a fetal pole without fetal heart rate (FHR) was in the lower segment of the uterus and cervix and the 2nd gestational sac (34 mm) was over the first one in the uterine cavity without a fetal pole. Two diagnoses were considered; the 2nd sac, and a low probability of sub chorionic hemorrhage, confirming a missed abortion. The 3rd ultrasound performed on 18 Nov, 2016 showed a gestational sac (23 mm, 7 to 8 weeks) with irregular margin and no fetal pole in the uterine cavity. Also, an echo-free region with irregular margin and echogenic debris (70 × 24 mm, 7 to 8 weeks) suggested the 2nd sac without a fetal pole or a clot in the lower part of the uterus (Figure 1). The increased local vascularity between one-third and two-thirds of superior portion of the lower part of the uterine body suggested either partial mole or a level of placenta accreta or trophoblastic reaction (Figure 2). No sonographers indicated the possibility of a CSP.

After getting patient's consent for hysterectomy, blood reservation, and prophylactic antibiotic, she received 400 mg vaginal misoprostol for cervical ripening. Patient's hemoglobin (Hb), hematocrit (HCT), and red blood cell (RBC) count were 10.4 g/dL, 30.2%, and 3.8 × 10^6 cells/μL, respectively. After 6 hours, curettage lasted 45 minutes under general anesthesia was performed in which plenty of tissues were extracted from the uterus. At the end of the surgery, severe bleeding occurred and continued and the uterine atony was notable. Then, 800 mg misoprostol plus methylergonovine and oxytocin was administered to induce uterine involution. Despite administration of medicine and fixing a Folly catheter in the uterus, bleeding continued.

In the next step, due to the continuation of bleeding and signs of hypovolemia (tachycardia, low blood pressure, and anuria), it was decided to perform a total abdominal hysterectomy. During the surgery, the patient received 5 units of packed RBCs, 4 units of fresh frozen plasma (FFP),

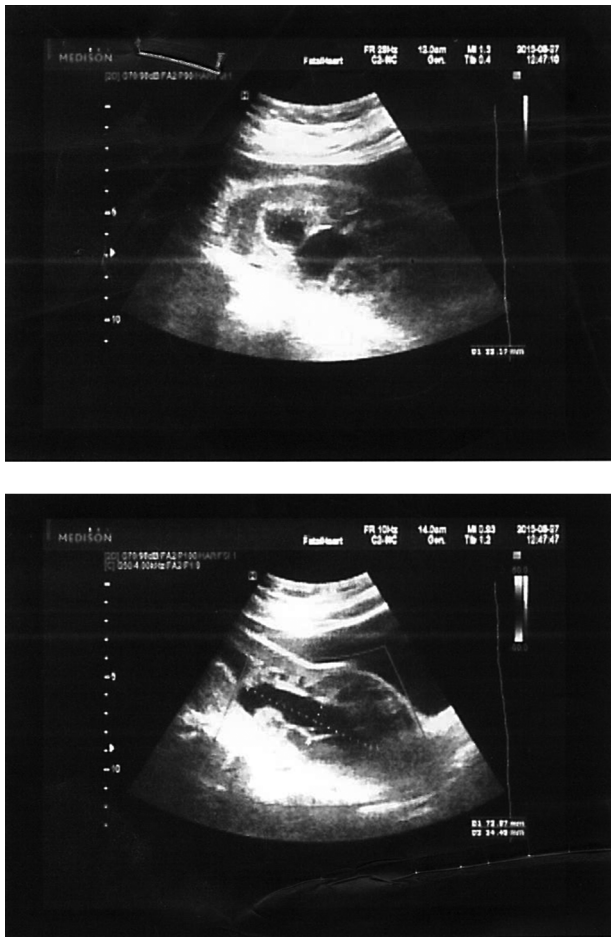

Figure 2. Transabdominal Images Suggesting a Gestation sac of 23 mm Diameter Developing in the Uterine Cavity (the 3rd Ultrasound)

3 units of cryoprecipitate, and 2 units platelet. Both ovaries and fallopian tubes remained. The lower segment of the uterus was very thin; placenta had been implanted in the lower segment of the uterus and still contained residue. The patient was discharged in good condition after 3 days with Hb 9.6 g/dL, HCT29.3%, and RBC 3.38 $\times$ 10^6 cells/μL; 10 days later sutures were removed and the patient had no problem.

3. Discussion

Although the probability of a CSP is increased due to increased cesarean and repeat cesarean rates, early accurate diagnosis of CSP is the key to perform proper treatment, especially a complicated CSP such as HCSP is a challenge. In the current case, the results of ultrasounds were confusing and the third-one suggested missed abortion. Both sacs had no fetal pole suggesting no need to preserve any sac in the uterus. After curettage and heavy bleeding, it was found that it was not a missed abortion.

Different approaches are suggested to manage CSP. Curettage at diagnosis with CSP is not recommended (7). Previous studies suggested systemic methotrexate treatment as the best approach, which can help the gestational sac to be bulged into the uterine cavity (8). Uterine artery embolization (UAE) (before uterine curettage) (9), hysteroscopy combined with UAE (10), and UAE combined with methotrexate followed by curettage (11) are other recommended approaches. A recent case report indicated that they could terminate CSP with 3 intra sac, intra muscular, and intra placental injections of methotrexate (12). Comparatively, in HCSP, researchers helped to terminate CSP and preserve intrauterine pregnancy. In a case report, researchers followed the HCSP with expectant management that resulted in the progress of the gestational sac at the cesarean scar site into the lower uterine cavity and continuation of development of both fetus, which resulted in full-term twin deliveries (13). In a case presented by Lincenberg, CSP ruptured uterus spontaneously and was expelled while the intrauterine fetus remained and developed in the uterus and was delivered by emergent cesarean (4). In another approach, the ectopic pregnancy was resected through mini laparotomy to conserve the intrauterine pregnancy (14). In another case report, expectant management was conducted and due to aggravation of CSP, selective fetal reduction of CSP was performed at 16 + 4 weeks of gestation, while the intrauterine pregnancy was preserved (15).

The current study reported a HCSP case. The weak point of the current study was that the diagnosis was made based on ultrasound findings. Two out of three ultrasound reports showed 2 gestational sacs and more tests were not requested to definitely confirm the diagnosis. The specific character in the current case was that HCSP occurred following a non-assisted pregnancy.

3.1. Conclusion

Due to the rising of cesarean and repeat cesarean incidences, and helps of diagnostic methods such as ultrasound, the incidence of CSP diagnosis is increasing. Furthermore, advances in reproductive technologies predict that the incidence of HSCP increases. Obstetricians and sonographers should consider CSP and HSCP as a differential diagnosis in females experiencing spotting and bleeding in the early stages of pregnancy with a history of cesarean delivery. HCSP does not have any specific symptoms and may be easily misdiagnosed. Physicians should use precise diagnostic tests such as transvaginal 3-dimensional color Doppler ultrasonography, hys-

teroscopy, and magnetic resonance imaging in the case of tests with controversial results and proper treatment to prevent hemorrhage or uterine rupture to preserve fertility.

Acknowledgments

Authors would like to thank the patient for her permission to publish the history of her present pregnancy.

Footnotes

Financial Disclosure: Authors declared no financial disclosure.

Ethical Considerations: Ethical approval for this report was not required, but the authors obtained a written consent from the patient for publication of the details and images.

Competing Interests: Authors declared no conflict of interest.

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