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

Mahsa Karbasi¹, Reza Aletaha¹, Ramin Ahangar-Sirous¹, Amir Honarmand Alamdari¹, esmaeil gharepapagh², and Sahar Rezaei¹

¹Tabriz University of Medical Sciences Faculty of Medicine ²Tabriz University of Medical Sciences

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Introduction

Heterotopic pregnancy (HP) is characterized by the existence of intrauterine and ectopic pregnancies occurring simultaneously(1). This is a rare vet 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 to 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, 7, 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, 10, 11). Due to the considerable risk for fetal and maternal morbidity and mortality, timely and precise diagnosis of HCSP is vital(12). The principal investigative approach that holds the greatest significance involves sonographic and Doppler flow-based evaluations(13). In the management of HCSP, preserving the coexistent presents a significant challenge. Guidelines for managing HCSP while preserving the IUP are not universally standardized due to the rarity of this condition (14). 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.

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 an 8-week 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.

Methods

The woman's lab results, which included a beta-human chorionic gonadotropin (β -hCG) measurement of 174,025 milli-international units/mL, were all found to be within the accepted range. Transvaginal ultra-

sonography (TVUS) revealed a dichorionic diamniotic twin gestation, with cardiac activity and crown-rump length appropriate for 7 weeks and 5 days of gestation. One of the gestational sacs was identified in the endometrial cavity while the other was found within the myometrium, specifically at the site of the earlier Cesarean scar, where the thickness of the myometrium measured 2.5 mm. (Figures 1,2) and rich blood flow (in color Doppler ultrasound examination) Amidst the gestational sac and the wall of the bladder. (Figure 3).

Results

The above findings were suggestive of a heterotopic Cesarean scar pregnancy. The patient and her husband received guidance regarding the management options and potential maternal and fetal complications associated with the continuation of the pregnancy, such as excessive bleeding, abortion, and the need for subsequent hysterectomy. After thorough counseling and as the couple did not want to preserve the intrauterine gestation, The obstetrician arranged for the patient to undergo an exploratory laparotomy coupled with a dilation and curettage (D & C) procedure. The surgery went well and bleeding during the surgery was about 500 ml. The ultrasonography demonstrated the complete evacuation of the uterus the following day and β -hCG was dropped to 68,344 milli-international units/mL 48 hours after the operation. The patient was discharged on postoperative day 3 and her next follow-ups were also unremarkable.

Discussion

HCSP is recognized as one of the least common forms of heterotopic pregnancies, requiring careful observation of a potential IUP (8, 15, 16). It has been documented that approximately 1 in 30,000 deliveries conventionally encompasses HPs. However, with the advent and continued use of ART, there has been an observable increase in the occurrence of HPs, which is currently approximated at 1% (15, 17). 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 (IVF) could occasionally lead to this outcome. There are multiple contributing factors that increase the probability of pregnancy in this location. These include frequent Cesarean sections that result in a larger scar area, as well as a breech presentation during a premeditated incision on an undeveloped lower uterine segment (12). CSP may either 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 the transvaginal ultrasound scan (TVUS) (18). 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 prior to rupture (19). 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 (20, 21, 22, 23). Moreover, A break in the front wall of the uterus can be unveiled via a sagittal view of the uterus, achieved when the ultrasound beam's direction crosses through the amniotic sac (24). In addition, color Doppler flow is a vital tool in accurately determining the implantation site (2). A major benefit of Doppler flow evaluations is the capacity to differentiate a viable pregnancy located in the scar region from a non-viable intrauterine pregnancy (25). This distinction undeniably influences the treatment strategy to be adopted. If an intrauterine pregnancy proves to be 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 (25). he gestational mass that is positioned within the scar region exhibits a low-impedance flow rate (pulsatility index <1) and a high speed (peak velocity >20 cm/s) (25). Various investigators have also noted that the blood flow's resistance index is below 0.5, in conjunction with a peak value ratio of the systolic-to-diastolic (S/D) flow being less than 3 (19). TVUS, when used in conjunction with color Doppler analysis, exhibits a diagnostic sensitivity of 85% for the detection of a CSP (26). Recently, some clinicians have begun utilizing Three-dimensional (3D) ultrasonography and 3D Power Doppler imaging (27). Based on their findings, employing multiplanar views in conjunction with 3D-rendered images can enhance diagnostic precision in such circumstances. In cases where diagnosing becomes intricate or challenging, Magnetic Resonance Imaging (MRI) may also prove to be advantageous (28). The handling of HCSP usually poses a sophisticated task, particularly in scenarios where the woman indicates a wish to preserve the current intrauterine embryo (29). At present, there is no universally recognized and established treatment protocol specifically for handling HP involving CSP. Various strategies are available for managing a HCSP, encompassing watchful waiting, medical intervention, and surgical termination. Successful cases of expectant management have been documented in medical literature. However, given the unfavorable prognosis for an uncomplicated full-term pregnancy, this approach is generally not advised (12). The current medical treatment options for CSPs include transvaginal embryo aspiration, injection of potassium chloride (KCl) or hyperosmolar glucose, and local or systemic methotrexate (MTX) treatments (29). MTX has been recognized as a potential treatment choice for managing ectopic pregnancies in numerous case studies, however, there's apprehension that its use in HPS cases alongside an intrauterine pregnancy might lead to teratogenic effects and fetal abnormalities (6). A handful of cases describe the treatment of HCSP with viable pregnancies via local injection of potassium chloride, a method traditionally employed for fetal reduction in multiple pregnancies. However, treatment with potassium chloride entails certain inherent risks, including a heightened likelihood of abdominal discomfort, miscarriage, excessive vaginal bleeding, preterm birth, further surgical intervention, as well as spontaneous rupture of the amniotic membranes and the ensuing onset of chorioamnionitis (6). The foremost surgical treatment alternative for ectopic CSP usually entails the explicit extraction of the ectopic mass situated at the site of the earlier Cesarean scar via various methods, such as open laparotomy, hysteroscopy, laparoscopy, or dilatation and curettage (30). Some scholars have suggested a surgical approach as the primary management strategy for a HCSP (14). Surgical extraction of the CSP serves as a practical method to stave off antenatal complications like vaginal bleeding and the re-growth of gestational tissue. In addition, pelviscopic removal may aid in strengthening the lower uterine segment. An open laparotomy is commonly favored since it offers superior surgical control due to an expanded operational field, thus enhancing the chances of effectively controlling any excessive bleeding (30). The current case arrived at our facility exhibiting mild symptoms, and a HCSP with one viable intrauterine embryo aged 7 weeks and 5 days was identified via ultrasound examination. The couple was thoroughly advised about the potential hazards and complexities of HCSP, in addition to the available strategies for its management. Given that the couple had no intention to maintain the normally implanted viable intrauterine pregnancy, and Resulting from the thin layer of myometrium that lies between the gestational sac and the wall of the bladder posing a significant risk of heavy bleeding, The choice was made to carry out an exploratory laparotomy with D & C on the patient. The operation was successfully carried out without severe blood loss, and the patient's recovery proceeded without incident.

Conclusion

The proper management of a heterotopic Cesarean scar pregnancy requires timely diagnosis through ultrasonography. Early diagnosis allows for immediate intervention to prevent complications such as uterine rupture or potentially lethal bleeding.

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Availability of data and materials

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Consent for publication

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Competing interests

No author states to have any conflicts of interest.

References

1. Yu H, Luo H, Zhao F, Liu X, Wang X. Successful selective reduction of a heterotopic cesarean scar pregnancy in the second trimester: a case report and review of the literature. BMC Pregnancy and Childbirth. 2016;16(1):1-7.

2. Ouyang Y, Chen H, Lin G, Xiang S, Qin J, Gong F, et al. Heterotopic Cesarean Scar Pregnancy: An Analysis of 20 Cases Following in vitro Fertilization-Embryo Transfer. Journal of Ultrasound in Medicine. 2021;40(10):2239-49.

3. Barrenetxea G, Barinaga-Rementeria L, de Larruzea AL, Agirregoikoa JA, Mandiola M, Carbonero K. Heterotopic pregnancy: two cases and a comparative review. Fertility and sterility. 2007;87(2):417. e9-. e15.

4. Goldberg JM, Bedaiwy MA. Transvaginal local injection of hyperosmolar glucose for the treatment of heterotopic pregnancies. Obstetrics & Gynecology. 2006;107(2 Part 2):509-10.

5. Dor J, Seidman DS, Levran D, Ben-Rafael Z, Ben-Shlomo I, Mashiach S. The incidence of combined intrauterine and extrauterine pregnancy after in vitro fertilization and embryo transfer. Fertility and sterility. 1991;55(4):833-4.

6. Vikhareva O, Nedopekina E, Herbst A. Normal vaginal delivery at term after expectant management of heterotopic caesarean scar pregnancy: a case report. Journal of Medical Case Reports. 2018;12:1-3.

7. OuYang Z, Yin Q, Xu Y, Ma Y, Zhang Q, Yu Y. Heterotopic cesarean scar pregnancy: diagnosis, treatment, and prognosis. Journal of Ultrasound in Medicine. 2014;33(9):1533-7.

8. Kim ML, Jun HS, Kim JY, Seong SJ, Cha DH. Successful full-term twin deliveries in heterotopic cesarean scar pregnancy in a spontaneous cycle with expectant management. Journal of Obstetrics and Gynaecology Research. 2014;40(5):1415-9.

9. Salomon L, Fernandez H, Chauveaud A, Doumerc S, Frydman R. Successful management of a heterotopic Caesarean scar pregnancy: potassium chloride injection with preservation of the intrauterine gestation: case report. Human Reproduction. 2003;18(1):189-91.

10. Timor-Tritsch IE, Monteagudo A. Unforeseen consequences of the increasing rate of cesarean deliveries: early placenta accreta and cesarean scar pregnancy. A review. American journal of obstetrics and gynecology. 2012;207(1):14-29.

11. Barnhart KT. Early pregnancy failure: beware of the pitfalls of modern management. Fertility and sterility. 2012;98(5):1061-5.

12. Maymon R, Halperin R, Mendlovic Se, Schneider D, Herman A. Ectopic pregnancies in a Caesarean scar: review of the medical approach to an iatrogenic complication. Human reproduction update. 2004;10(6):515-23.

13. Marchiole P, Gorlero F, De Caro G, Podestà M, Valenzano M. Intramural pregnancy embedded in a previous Cesarean section scar treated conservatively. Ultrasound in Obstetrics and Gynecology: The Official Journal of the International Society of Ultrasound in Obstetrics and Gynecology. 2004;23(3):307-9.

14. Chen Z-Y, Zhou Y, Qian Y, Luo J-M, Huang X-F, Zhang X-M. Management of heterotopic cesarean scar pregnancy with preservation of intrauterine pregnancy: A case report. World Journal of Clinical Cases. 2021;9(22):6428.

15. Ugurlucan FG, Bastu E, Dogan M, Kalelioglu I, Alanya S, Has R. Management of cesarean heterotopic pregnancy with transvaginal ultrasound–guided potassium chloride injection and gestational sac aspiration, and review of the literature. Journal of Minimally Invasive Gynecology. 2012;19(5):671-3.

16. Lincenberg KR, Behrman ER, Bembry JS, Kovac CM. Uterine rupture with cesarean scar heterotopic pregnancy with survival of the intrauterine twin. Case reports in obstetrics and gynecology. 2016;2016.

17. Lee JS, Cha H-H, Han AR, Lee SG, Seong WJ. Heterotopic pregnancy after a single embryo transfer. Obstetrics & gynecology science. 2016;59(4):316-8.

18. Timor-Tritsch IE, Monteagudo A, Cali G, El Refaey H, Agten AK, Arslan AA. Easy sonographic differential diagnosis between intrauterine pregnancy and cesarean delivery scar pregnancy in the early first trimester. American journal of obstetrics and gynecology. 2016;215(2):225. e1-. e7.

19. Weimin W, Wenqing L. Effect of early pregnancy on a previous lower segment cesarean section scar. International Journal of Gynecology & Obstetrics. 2002;77(3):201-7.

20. Godin P-A, Bassil S, Donnez J. An ectopic pregnancy developing in a previous caesarian section scar. Fertility and sterility. 1997;67(2):398-400.

21. Seow K-M, Cheng W-C, Chuang J, Lee C, Tsai Y-L, Hwang J-L. Methotrexate for cesarean scar pregnancy after in vitro fertilization and embryo transfer. A case report. The Journal of reproductive medicine. 2000;45(9):754-7.

22. Seow KM, Huang LW, Lin YH, Yan-Sheng Lin M, Tsai YL, Hwang JL. Cesarean scar pregnancy: issues in management. Ultrasound in Obstetrics and Gynecology: The Official Journal of the International Society of Ultrasound in Obstetrics and Gynecology. 2004;23(3):247-53.

23. Fylstra DL. Ectopic pregnancy within a cesarean scar: a review. Obstetrical & gynecological survey. 2002;57(8):537-43.

24. Vial Y, Petignat P, Hohlfeld P. Pregnancy in a cesarean scar. Ultrasound in Obstetrics and Gynecology: The Official Journal of the International Society of Ultrasound in Obstetrics and Gynecology. 2000;16(6):592-3.

25. Jurkovic D, Hillaby K, Woelfer B, Lawrence A, Salim R, Elson C. First-trimester diagnosis and management of pregnancies implanted into the lower uterine segment Cesarean section scar. Ultrasound in Obstetrics and Gynecology: The Official Journal of the International Society of Ultrasound in Obstetrics and Gynecology. 2003;21(3):220-7.

26. Rotas MA, Haberman S, Levgur M. Cesarean scar ectopic pregnancies: etiology, diagnosis, and management. Obstetrics & Gynecology. 2006;107(6):1373-81.

27. Shih JC. Cesarean scar pregnancy: diagnosis with three-dimensional (3D) ultrasound and 3D power Doppler. Ultrasound in Obstetrics and Gynecology: The Official Journal of the International Society of Ultrasound in Obstetrics and Gynecology. 2004;23(3):306-7.

28. Duenas-Garcia OF, Young C. Heterotopic cesarean scar pregnancy associated with a levonorgestrelreleasing intrauterine device. International Journal of Gynaecology and Obstetrics: the Official Organ of the International Federation of Gynaecology and Obstetrics. 2011;114(2):153-4. 29. Demirel LC, Bodur H, Selam B, Lembet A, Ergin T. Laparoscopic management of heterotopic cesarean scar pregnancy with preservation of intrauterine gestation and delivery at term: case report. Fertility and Sterility. 2009;91(4):1293. e5-. e7.

30. Kim H, Koh JH, Lee J, Sim Y, Lee S-H, Lee S-J, et al. 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. Diagnostics. 2022;12(3):762.

Legends

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 TVUS 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).

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

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