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Contents

Thrice Monthly Volume 11 Number 17 June 16, 2023

REVIEW

3932 Liver replacement therapy with extracorporeal blood purification techniques current knowledge and future directions

Papamichalis P, Oikonomou KG, Valsamaki A, Xanthoudaki M, Katsiafylloudis P, Papapostolou E, Skoura AL, Papamichalis M, Karvouniaris M, Koutras A, Vaitsi E, Sarchosi S, Papadogoulas A, Papadopoulos D

MINIREVIEWS

3949 Prediction models for recurrence in patients with small bowel bleeding

Kim JH, Nam SJ

3958 Investigation of possible relationship between atopic dermatitis and salivary biomarkers, stress, and sleep disorders

Estefan J, Ferreira DC, Cavalcante FS, dos Santos KRN, Ribeiro M

- Value of clinical applications of differential pressure and relative pressure imaging in the left ventricle 3967 Zheng AS, Yu HX
- 3976 Low-dose immunotherapy as a potentiator to increase the response with neo-adjuvant chemotherapy in oral cancers

Rathinasamy N, Muthu S, Krishnan A

3980 Kidney disease in patients with chronic liver disease: Does sex matter? Cooper KM, Colletta A, Moulton K, Ralto KM, Devuni D

ORIGINAL ARTICLE

Case Control Study

3993 Elabela is a reliable biomarker for predicting early onset preeclampsia: A comparative study Amer Ali E, Nori W, Salman AF, Al-Rawi TSS, Hameed BH, Al-Ani RM

Retrospective Cohort Study

4003 Acute-on-chronic liver failure is independently associated with higher mortality for cirrhotic patients with acute esophageal variceal hemorrhage: Retrospective cohort study

Terres AZ, Balbinot RS, Muscope ALF, Longen ML, Schena B, Cini BT, Rost Jr GL, Balensiefer JIL, Eberhardt LZ, Balbinot RA, Balbinot SS, Soldera J

Retrospective Study

4019 Elastic fiber degradation in the development of pediatric granuloma annulare: Report of 39 cases Zhang DY, Zhang L, Yang QY, Xie YC, Jiang HC, Li JZ, Shu H



6	World Journal of Clinical Cases				
Conten	Thrice Monthly Volume 11 Number 17 June 16, 2023				
4026	Anti-bacterial mechanism of baicalin-tobramycin combination on carbapenem-resistant <i>Pseudomonas</i> aeruginosa				
	Jin LM, Shen H, Che XY, Jin Y, Yuan CM, Zhang NH				
	SYSTEMATIC REVIEWS				
4035	Acknowledging the use of botanicals to treat diabetic foot ulcer during the 21st century: A systematic review				
	Narzary I, Swarnakar A, Kalita M, Middha SK, Usha T, Babu D, Mochahary B, Brahma S, Basumatary J, Goyal AK				
	CASE REPORT				
4060	Pregabalin induced balance disorder, asthenia, edema, and constipation in an elderly adult: A case report				
	Ma LP, Wen C, Zhao TX, Jiang XM, Gu J				
4065	Emergency internal iliac artery temporary occlusion after massive hemorrhage during surgery of cesarean scar pregnancy: A case report				
	Xie JP, Chen LL, Lv W, Li W, Fang H, Zhu G				
4072	Hemophagocytic lymphohistiocytosis after autologous stem cell transplantation in angioimmunoblastic T- cell lymphoma: A case report				
	Zhang ZR, Dou AX, Liu Y, Zhu HB, Jia HP, Kong QH, Sun LK, Qin AQ				
4079	Successful reconstruction of an ankle defect with free tissue transfer in a hemophilia A patient with repetitive hemoarthrosis: A case report				
	Lee DY, Lim S, Eo S, Yoon JS				
4084	Primary pelvic Echinococcus granulosus infection: A case report				
	Abulaiti Y, Kadi A, Tayier B, Tuergan T, Shalayiadang P, Abulizi A, Ahan A				
4090	Epstein-Barr virus-induced infection-associated hemophagocytic lymphohistiocytosis with acute liver injury: A case report				
	Sun FY, Ouyang BQ, Li XX, Zhang T, Feng WT, Han YG				
4098	Cardiac arrest secondary to pulmonary embolism treated with extracorporeal cardiopulmonary resuscitation: Six case reports				
	Qiu MS, Deng YJ, Yang X, Shao HQ				
4105	Flared inflammatory episode transforms advanced myelodysplastic syndrome into aplastic pancytopenia: A case report and literature review				
	Ju B, Xiu NN, Xu J, Yang XD, Sun XY, Zhao XC				
4117	Frontal penetrating arrow injury: A case report				
	Rodríguez-Ramos A, Zapata-Castilleja CA, Treviño-González JL, Palacios-Saucedo GC, Sánchez-Cortés RG, Hinojosa- Amaya LG, Nieto-Sanjuanero A, de la O-Cavazos M				
4123	Chest wall osteochondroma resection with biologic acellular bovine dermal mesh reconstruction in pediatric hereditary multiple exostoses: A case report and review of literature				
	Alshehri A				



.	World Journal of Clinical Cases
Conten	Thrice Monthly Volume 11 Number 17 June 16, 2023
4133	Massive pulmonary embolism in Klippel-Trenaunay syndrome after leg raising: A case report
	Lo CY, Chen KB, Chen LK, Chiou CS
4142	Improved super-elastic Ti-Ni alloy wire intrusion arch for skeletal class II malocclusion combined with deep overbite: A case report
	Yang CY, Lin CC, Wang IJ, Chen YH, Yu JH
4152	Glucocorticoid pulse therapy in an elderly patient with post-COVID-19 organizing pneumonia: A case report
	Park S, Jang Y, Koo SM, Nam BD, Yoon HY
4159	Endoscopic and surgical treatment of jejunal gallstone ileus caused by cholecystoduodenal fistula: A case report
	Fan WJ, Liu M, Feng XX
4168	Application of advanced platelet-rich fibrin for through-and-through bony defect during endodontic surgery: Three case reports and review of the literature
	Algahtani FN, Almohareb R, Aljamie M, Alkhunaini N, ALHarthi SS, Barakat R
4179	Facial Merkel cell carcinoma in a patient with diabetes and hepatitis B: A case report
	Ren MY, Shi YJ, Lu W, Fan SS, Tao XH, Ding Y
4187	Pregnancy and lactation-associated osteoporosis with pyogenic spondylitis: A case report
	Zhai K, Wang L, Wu AF, Qian Y, Huang WM
4194	Hourglass-like constriction of the anterior interosseous nerve in the left forearm: A case report
	He R, Yu JL, Jin HL, Ng L, Wang JC, Li X, Gai TT, Zhou Y, Li DP
4202	Crohn's disease in human immunodeficiency virus-infected patient: A case report
	Vinikaite A, Kurlinkus B, Jasinskaite D, Strainiene S, Buineviciute A, Sadauskaite G, Kiudelis V, Kazenaite E



Contents

Thrice Monthly Volume 11 Number 17 June 16, 2023

ABOUT COVER

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CASE REPORT

Emergency internal iliac artery temporary occlusion after massive hemorrhage during surgery of cesarean scar pregnancy: A case report

Ji-Ping Xie, Lin-Lin Chen, Wen Lv, Wu Li, Hui Fang, Guang Zhu

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Abstract

BACKGROUND

Cesarean scar pregnancy (CSP) is rare but may result in uterine rupture during pregnancy or massive hemorrhage during abortion procedures. Awareness of this condition is increasing, and most patients with CSP are now diagnosed early and can be managed safely. However, some atypical patients are misdiagnosed, and their surgical risks are underestimated, increasing the risk of fatal hemorrhage.

CASE SUMMARY

A 27-year-old Asian woman visited our institution because of abnormal pregnancy, and she was diagnosed with a hydatidiform mole through transvaginal ultrasound (TVS). Under hysteroscopy, a large amount of placental tissue was found in the scar of the lower uterine segment, and a sudden massive hemorrhage occurred during the removal process. The bilateral internal iliac arteries were temporarily blocked under laparoscopy, and scar resection and repair were rapidly performed. She was discharged in good condition 5 d after the operation.

CONCLUSION

Although TVS is widely used in the diagnosis of CSP, delays in the diagnosis of atypical CSP remain. Surgical treatment following internal iliac artery temporary occlusion may be an appropriate management method for unanticipated massive hemorrhage during CSP surgery.

Key Words: Internal iliac artery temporary occlusion; Cesarean scar pregnancy; Uterine artery embolization; Misdiagnosis; Hysteroscopy; Laparoscopy; Case report



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Core Tip: Unanticipated massive hemorrhage during cesarean scar pregnancy surgery can lead to serious complications. If not effectively managed, it can lead to hemorrhagic shock or even death. Uterine artery embolization is usually used to prevent massive hemorrhage, but it requires multidisciplinary cooperation and cannot be performed immediately in the case of emergency. We adopted bilateral internal iliac artery emergency temporary occlusion to control intraoperative bleeding immediately without postoperative complications, and this may be a feasible method.

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INTRODUCTION

The term "cesarean scar pregnancy" (CSP) describes pregnancies that develop after the implantation of fertilized eggs, embryos, or gestational sacs at the site of a prior cesarean delivery scar[1]. CSP occurs in 4 percent of all pregnancies, with a reported frequency of 1/2216-1/1800[2,3]. Cesarean section (CS) rates have been rising over the world in recent years, and in China, the relaxation of the two-child policy may increase the additional risk of CSP in reproductive-aged women with a history of CS[4]. Delaying CSP therapy might result in major side effects such as bleeding, uterine rupture, hysterectomy, and possibly loss of fertility[5]. Thus, the standard management of CSP is the timely termination of pregnancy.

Transvaginal ultrasound (TVS) is the most effective tool and first-line imaging modality for diagnosing CSP[6,7]; early diagnosis and prompt treatment are vital for reducing maternal mortality. Unfortunately, the research reports a shockingly high number of missed diagnoses of CSP, 107 (13.6%) of the 751 instances of CSP were overlooked or misdiagnosed, which increased morbidity[8]. Therefore, identifying methods to manage CSP in an emergency to reduce the occurrence of complications and protect the fertility of patients is crucial. In this paper, we report a case of a patient with CSP that was misdiagnosed by TVS as a hydatidiform mole who suffered massive hemorrhage during hysteroscopic surgery, underwent laparoscopic scar resection and repair after internal iliac artery temporary occlusion, and had a positive outcome. The Tongde Hospital's Institutional Review Board verified that ethical approval of this retrospective study was exempt.

CASE PRESENTATION

Chief complaints

A 27-year-old Chinese woman presented to the outpatient department of our hospital with abnormal pregnancy.

History of present illness

The patient had experienced menopause for 60 d. Her normal menstrual cycle was 27-35 d, and a urine pregnancy test was positive at 40 d of menopause. After 52 d of menopause, 174.8 mIU/mL of blood beta human chorionic gonadotropin (β -hCG) and 3.79 nmol/L of progesterone were examined in the hospital, and TVS indicated a honeycomb mixed echo in the uterine cavity of approximately 7.6 cm × 6.2 cm and a rich blood flow, some of which seemed to be embedded in the CS scar. The patient had no abdominal pain or vaginal bleeding and was experiencing a mild early pregnancy reaction. She was admitted to our hospital because of the abnormal pregnancy indicated by TVS.

History of past illness

The patient had been delivered by CS due to macrosomia 4 years previously.

Personal and family history

The patient has no relevant family history.



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Figure 1 Transvaginal ultrasound of cesarian scar pregnancy. A and B: White arrows indicate gestational matter (A) and dilated blood vessels (B) at the uterine scar

Physical examination

The patient had a temperature of 36.7 °C, heart rate of 82 bpm, respiratory rate of 20 breaths per minute, and blood pressure of 117/62 mmHg, and the oxygen saturation in room air was 99%. After admission, a gynecological examination revealed some bloody secretion in the vagina, mild cervical erosions without lifting pain, enlargement of the anterior uterus similar to that at 60 d of pregnancy without tenderness, and no abnormalities in the bilateral appendages.

Laboratory examinations

Hemoglobin level was 126 g/L, blood β -hCG was 189.6 mIU/mL, progesterone was 1.0 nmol/L, and the white cell and platelet count were normal. The prothrombin and partial thromboplastin times were normal, and the d-dimer value was 1.98 mg/L. Serum C-reactive protein was normal. The blood biochemistry and urine analyses were normal.

Imaging examinations

According to the TVS review, the scar area revealed a disorderly high echo mass, and multiple vesicular echoes were found in utero, with the largest being approximately 2.2 cm × 1.1 cm (Figure 1). An embryo abortion was considered, and a hydatidiform mole could not be excluded. The electrocardiogram and chest computed tomography scan were also normal.

FINAL DIAGNOSIS

The final diagnosis of the presented case was unclear, with CSP, a hydatidiform mole, and spontaneous abortion all considered.

TREATMENT

Hysteroscopic surgery with informed consent was planned for further diagnosis. Through hysteroscopy, we observed that a large amount of tissue with blood clots had adhered to the scar in the lower segment of the uterine cavity, and no obvious pregnancy sac or hydatidiform mole tissue was identified. When we clamped the scar tissue and blood clots, a large amount of blood flowed from the uterine cavity and was difficult to control. We used two vascular forceps to rapidly clamp the bilateral uterine arteries from the cervix and the cervix itself, thus reducing the bleeding, and laparoscopy was performed. During laparoscopic surgery, we observed that the uterus was enlarged to a size similar to 60 d after pregnancy. In addition, the lower scar was obviously enlarged, and the blood vessels on the surface were clearly dilated. The posterior peritoneum was opened and the bilateral internal iliac arteries dissociated. A no. 1 absorbable suture was used as a slipknot to temporarily stop the blood flow (Figure 2). The scar was then incised, pregnancy tissue removed, scar cutting edge trimmed, and uterine wall repaired (Figure 3). Finally, hysteroscopy was performed to examine the uterine cavity, with no pregnancy tissue being found or diverticula formed. To confirm that there was no obvious bleeding on the suture surface, the internal iliac artery was untied to restore blood flow. The uterine wound was checked for bleeding after the pelvic cavity was cleaned with warm saline and diluted iodophor. We



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Figure 2 Intraoperative management of the internal iliac artery. A: Temporary occlusion of the internal iliac artery; B: Release of the temporary occlusion of the internal iliac artery.



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Figure 3 Pregnancy tissue removed and uterine scars repaired. A: Removal of pregnancy tissue with spoon forceps through the vagina; B: Repair of the myometrium of the anterior uterine wall after scar trimming.

> stitched up the wound afterward. Our patient got two units of packed red blood cells throughout the procedure, and there were no acute issues despite an estimated intraoperative blood loss of 1000 mL.

OUTCOME AND FOLLOW-UP

Postoperatively, we used cefuroxime sodium to prevent infection, oral iron for erythropoiesis, and oxytocin to promote uterine contractions. We observed that the β -hCG was 35.2 mIU/mL on postoperative day 1, and 3 d following surgery, and the usual inflammatory signs progressively returned to normal levels. A routine blood examination suggested mild anemia and no obvious abnormalities in the blood biochemistry or coagulation function. On the first postoperative day, the patient anal exhaled, and on the third postoperative day, the patient defecated. During hospitalization, she experienced some vaginal bleeding but no symptoms such as abdominal pain, fever, thrombosis, or intestinal obstruction. The patient recovered well and was discharged on postoperative day 5.

Pathological report: Vestigial villi and decidual tissue were observed in the scar tissue of the uterus. During the follow-up assessment approximately 1 wk after discharge, the patient's β -hCG levels had returned to normal (2.9 mIU/mL), and 1 mo after discharge, a vaginal three-dimensional ultrasound indicated that the blood supply to the uterus and ovaries was normal.

DISCUSSION

CSP is a rare and iatrogenic form of ectopic pregnancy that is considered a potentially life-threatening



condition. It is crucial to diagnose problems quickly and treat them right away. An empty endometrial cavity and cervical canal, the presence of a gestational sac in the anterior uterine wall, and a strong trophoblastic/placental blood flow are all factors that aid in the diagnosis of CSP through transvaginal ultrasonography[9-11]. Studies have reported that the accuracy of TVS in diagnosing CSP is only 84.6% [2], with some misdiagnosis or missed diagnosis known to occur. The diagnosis accuracy of TVS depends on the pregnancy period. In continuing pregnancies, after 7 wk, the gestational sac migrates toward the fundus populating the uterine cavity. If the patient's previous history of CS and scar site examination are ignored, clinicians are often misled into believing that it is a normal intrauterine pregnancy[6]; incomplete abortion with an intrauterine hematoma could also affect the doctor's judgment of CSP, resulting in misdiagnosis. However, the inaccuracy of diagnosis directly affects the quality of the subsequent treatment, increasing the risk of hemorrhage and morbidity.

Our patient was misdiagnosed through TVS as having a hydatidiform mole, which may be related to abnormally high vascularities and varicosities in the anterior lower segment of the uterus. In addition, the low levels of β -hCG may explain why clinicians did not diagnose CSP, as β -hCG levels usually represent the activity of placental trophoblastic cells. However, studies have demonstrated that even at low levels of β -hCG, when the blood vessels are exposed after the pregnancy tissue has been cleared, contractions of the muscle layer cannot effectively stop bleeding in defects in the muscle layer at the scar site, leading to the occurrence of unpredicted catastrophic hemorrhage. Therefore, for patients with uterine scar, early ultrasound examination findings are key, even if the patient has a miscarriage or low β -hCG levels. However, this does not mean that the risk of surgery is low, as this case illustrates. Only by formulating a complete surgical program can the risk of severe postoperative complications be reduced.

The ideal therapy for CSP would include the safe removal of pregnant tissue, the repair of the uterine abnormality, and the maintenance of fertility. According to published reports, contemporary treatment options include medication, uterine artery embolization (UAE) in conjunction with hysteroscopic resection, dilatation and curettage, laparotomy, laparoscopy, and transvaginal resection[12]. UAE is suitable for patients with refractory CSP at high risk of massive hemorrhage and can be combined with all the aforementioned surgical methods to effectively prevent bleeding. Injury to the ovarian and urinary systems, as well as rectal perforation, sepsis, and pulmonary embolism, are all possible outcomes of UAE[13-16]. In addition, the operation requires interdisciplinary cooperation, primarily for preoperative planning, but it is difficult to quickly complete patient transport and preparation for interventional surgery when sudden massive bleeding is encountered.

The characteristic of this case is the occurrence of unpredicted vaginal hemorrhage during the operation. Under laparoscopy, the blood vessels on the surface of the scar were obviously dilated; if the scar had been removed, the uterine hemorrhage would have been aggravated. UAE may not be available in the case of emergency, and if it is performed, the type of non-predictable emergency operation required may result in postoperative complications. In addition, the patient was young and wanted her fertility to be preserved, which limited the plan for hysterectomy. In this case, through bilateral internal iliac artery temporary occlusion, the hemostatic effect during the operation was satisfactory, the residual pregnancy contents in utero were removed, and the uterine scar was repaired, which could improve the patient's reproductive outcome. The method was simple and easy to operate, and blood supply was restored immediately after the removal of the blockage. No postoperative complications were noted, the postoperative hospital stay was shortened, and the medical expenses were reduced.

The earliest treatment of CSP after laparoscopic bilateral internal iliac artery ligation was reported in 2006[17]. By temporarily blocking off both internal iliac arteries, blood flow to the uterus may be reduced by 48%, and pulse pressure can be reduced by 85% [18], resulting in better control of bleeding than with UAE because of vascular anastomosis between the internal iliac artery and its three offshoots (the uterine artery, the vaginal artery, and the internal pudendal artery). According to a review of recent literature[12,19-21], bilateral internal iliac artery temporary occlusion seems to be an effective strategy for reducing bleeding during CSP treatment without causing massive bleeding; hysteroscopy is required to deal with intrauterine lesions. Because the internal iliac artery occlusion is temporary, no serious complications are observed (Table 1). This method is also suitable for acute intraoperative bleeding and is superior to UAE^[20] and hysterectomy. However, it may have defects because a high level of surgical skill is required. In addition, few cases have been reported, and some complications may not have been detected. In order to confirm the efficacy and safety of this method, bigger sample size studies are necessary.

CONCLUSION

Although the misdiagnosis of CSP is rare, it can lead to serious complications, resulting in lifethreatening hemorrhage. A safe and simple treatment is needed for accidental massive bleeding during CSP surgery, and bilateral internal iliac artery temporary occlusion may be a viable treatment option, but more cases and multicenter cohort studies are required to verify this conclusion.



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Table 1 Summary of literature on temporary occlusion of internal iliac artery in the treatment of cesarean scar pregnancy since 2018							
	Li e <i>t al</i> [<mark>19</mark>], 2018	Xu et al <mark>[12</mark>], 2019	Zhao e <i>t al</i> [<mark>20</mark>], 2022	Su e <i>t al</i> [<mark>21</mark>], 2022			
Number of participants	1	5	33	21			
CSP type	ш	П	III	Ш			
Age (yr)	28	28-39	28.97 ± 4.61	34.38 ± 4.30			
Menelipsis time (d)	77	45-60	48.51 ± 2.39	47.35 ± 12.73			
Blood β -hCG before operation (mIU/mL)	40542	4569-67762	18906.12 ± 11296.74	2.62 ± 0.76 (10000 mol/L)			
Operation time (min)	85	60-165	126.47 ± 13.21	Not described			
Occlusion time (min)	27	20-26	Not described	30 ± 12.05			
Intraoperative blood loss (mL)	Minimal	20-40	112.44 ± 22.65	67.14 ± 32.78			
Blood transfusion	No	No	No	No			
Decrease rate of serum β -hCG 24 h after surgery (%)	Not described	Not described	81 ± 2.15	Not described			
Length of hospital stay (d)	3	2-3	5.28 ± 1.21	5.14 ± 0.32			
Incidence of postoperative complications (%)	None	None	6 ± 2.320	None			
Time to β -hCG normalization (wk)	Not described	3-4	Not described	Not described			
Ovarian function 3-6 mo after surgery	Not described	Not described	Normal	Not described			

β-hCG: Beta human chorionic gonadotropin; CSP: Cesarean scar pregnancy.

FOOTNOTES

Author contributions: Xie JP reviewed the literature and contributed to manuscript drafting; Chen LL involved in the data curation; Lv W supervised the findings of this work; Li W did the surgical appraisal; Fang H analyzed and interpreted the imaging findings; Zhu G was responsible for the revision of the manuscript for important intellectual content; and all authors issued final approval for the version to be submitted.

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