

Heterotopic Cesarean Scar Pregnancy in a Spontaneous Cycle

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ABSTRACT

Introduction: We report a viable cesarean scar pregnancy with a coexistent intrauterine pregnancy, managed successfully with operative hysteroscopy and suction curettage.

Case Description: A 32-year-old woman, with previous cesarean delivery, presented with vaginal bleeding and was referred to our hospital at 9 weeks' gestation for heterotopic pregnancy in a spontaneous cycle. An intrauterine pregnancy combined with a cesarean scar pregnancy was shown by ultrasonography. The patient underwent hysteroscopy-directed evacuation of the cesarean scar pregnancy and suction curettage with good hemostasis.

Discussion: Spontaneous heterotopic cesarean scar pregnancy is a rare but possible entity. The combination of hysteroscopy and suction curettage is an option for a minimally invasive procedure for heterotopic cesarean scar pregnancy.

Key Words: Cesarean scar, Heterotopic, Hysteroscopy, Pregnancy.

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INTRODUCTION

Heterotopic pregnancies (HPs) involving gestational tissue implanted in the endometrial cavity and extraendometrial cavity simultaneously are rare in the general population, with a range of occurrence estimated between 1 in 7 963 and 1 in 30 000.^{1,2} With the increasing use of assisted reproductive technologies, the incidence of HP is estimated to be as high as 0.2% to 1%.³ According to a review of the published literature from 1994 to 2004, most of the HPs were located in the fallopian tube (72.5%) and 3.8% were located on cesarean scars.⁴ Heterotopic cesarean scar pregnancy (HCSP) is even more unusual in a spontaneous cycle.

Management of cesarean scar pregnancy (CSP) itself carries risk of uncontrollable bleeding, not to mention the salvage of the

coexisting intrauterine pregnancy. In a previous report, we demonstrated successful conservative management of HCSP after in vitro fertilization–embryo transfer and preservation of intrauterine pregnancy.⁵ We report a case of HCSP in a spontaneous cycle, managed successfully by hysteroscopy-assisted suction curettage. In addition, a review of the published literature on spontaneous HCSPs diagnosed and treated from January 1992 to January 2012 is presented.

CASE REPORT

A 32-year-old Taiwanese woman, gravida 4, para 2, presented with vaginal bleeding. Her second infant was delivered by transverse lower-segment cesarean section 2 years earlier. The patient was referred to our hospital at 9 weeks'

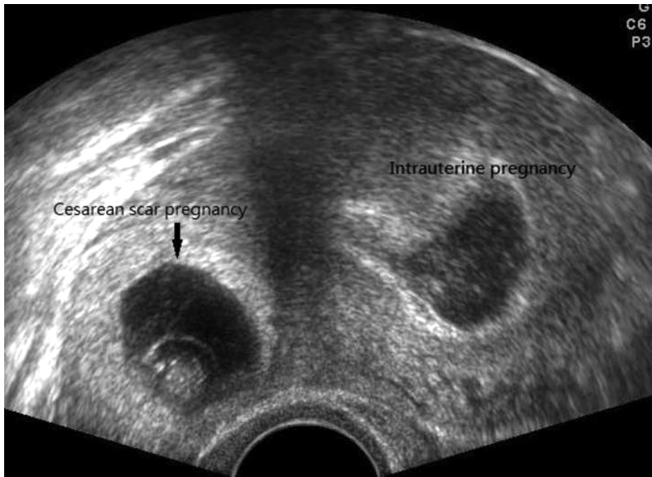


Figure 1. An ultrasonographic image shows a heterotopic twin pregnancy with one gestational sac located in the lower portion of the uterine cavity and the other within the cesarean section scar.

gestation for sonographic suspicion of a cervical pregnancy. Transvaginal sonography showed 2 gestational sacs, 1 located in the uterine lower portion without fetal cardiac activity and 1 located in the previous cesarean scar (**Figure 1**). The sac within the previous cesarean scar contained a yolk sac and a viable embryo measuring 8 weeks 2 days of gestation. Both ovaries appeared normal, and there were no adnexal masses or free fluid in the cul-de-sac. The plasma β human chorionic gonadotropin level was 68 500 mIU/mL, and general physical examination findings were normal. A diagnosis of HCSP was highly suspected. The woman did not desire to continue the pregnancy. After counseling, the couple opted for conservative treatment with operative hysteroscopy and suction curettage.

Under general anesthesia without endotracheal intubation, the patient was placed in the dorsolithotomy position. After a speculum was placed inside the vagina, a tenaculum was applied to the cervix and gentle traction was exerted to align the uterus. The cervix was dilated by Hegar dilators to 12 mm, and a continuous-flow 26F hysteroscopic resectoscope (Karl Storz, Tuttlingen, Germany) with a 90° wire loop electrode was introduced under ultrasonographic control. Uterine distension was achieved with distilled water propelled by simple gravity. An Aspen Excalibur electro-surgical generator (Aspen Labs, Englewood, Colorado) was used at a setting of 80 W of cutting waveform current and 100 W of coagulation current.

The intervention began by identifying the implantation of the ectopic sac. The electric loop of the resectoscope was used to push the gestational sac, exposing the vessel bed

of the implantation site. A coagulation current of 100 W was used for hemostasis. A placenta forceps was then used to pull out the gestational sacs under sonographic guidance, and suction curettage was used to clear the residual gestational tissue. Finally, a hysteroscopic rolling ball was used to stop the bleeding point. The operating time was 16 minutes. Vaginal bleeding was minimal at the end of the procedure. Postoperative ultrasonography showed a normal anteverted uterus with an intact endometrium and complete disappearance of the ectopic gestational sac. The patient had an unremarkable postoperative course and was discharged 3 hours after the procedure. A urine pregnancy test was negative on the 30-day postoperative visit, and normal echotexture of the uterus was noted. Menstruation resumed 4 days after this visit, and there has been no subsequent abnormal uterine bleeding within the 4-month follow-up period.

DISCUSSION

The exact incidence rate of HCSP is hard to estimate. A total of 19 878 pregnancies (unwanted pregnancies not included) were delivered and 846 ectopic pregnancies received treatment in our department from January 2007 to December 2011. Among these cases, 3 patients were diagnosed with HCSP and 1 of these 3 patients conceived spontaneously. Therefore the incidence rate of HCSP was at least 0.005% (1 of 20 724) at our hospital (a 3 900-bed, large medical center).

We performed a review of the published literature from January 1992 to January 2012 using the PubMed database. The medical terms used were *heterotopic pregnancy*, *cesarean scar pregnancy*, and *cervical pregnancy*. Available data on the patients' clinical characteristics, treatment, and outcome are summarized in **Table 1**. Our previous case and current case were included.

Ultrasonography is the primary imaging technique for diagnosing HCSP. The sonographic criteria for HCSP on a sagittal view of the uterus are as follows: a gestational sac with or without cardiac activity located anteriorly at the level of the internal os, equivalent to a prior lower uterine segment with a cesarean section scar; a bulging gestational sac with a diminished myometrial layer between the bladder and the sac; and the other gestational sac located in the normal endometrial cavity.^{5,6} The gestational sac in a spontaneous abortion in progress is avascular; therefore Doppler ultrasonography can provide more detail on the trophoblastic circulation of implantation to make a prompt differential diagnosis.⁷

At present, there is no standard treatment protocol for HCSP. Feticide with potassium chloride (KCl) under trans-

Table 1. Review of English-Language Literature Describing Successful Management of Heterotopic Cesarean Scar Pregnancy in Spontaneous Cycle

Reference	Case No.	Age, y	Gravity/Parity	Previous CS, ^a n	Gestation, wk	Fetal Heartbeat	Preoperative β -hCG ^a Level, mIU/mL	Management	Operating Time, min	Postoperative Stay, h	Outcome
Jurkovic et al, ⁸ 2003	1	36	10/3	3	7	+	68 800	TVS ^a guidance, potassium chloride-directed injection	NK ^a	NK	Emergency CS at 31 wk because of major antepartum hemorrhage
Porpora et al, ⁹ 2003	2	29	4/1	1	6	+	47 813	Sonographically guided aspiration	NK	144	Intended to preserve intrauterine pregnancy; fetal death on postoperative day 1
Kumar et al, ¹⁰ 2004	3	32	2/1	1	7	+	NK	TVS guidance, potassium chloride-directed injection	NK	24	β -hCG became negative 35 days after procedure
Yazicioglu et al, ¹¹ 2004	4	23	2/1	1	6	+	NK	TVS guidance, potassium chloride-directed injection	NK	NK	CS at 35 wk due to eclampsia; ligation of anterior division of both internal iliac arteries because of hemorrhage; live newborn
Demirel et al, ¹² 2009	5	34	2/1	1	6	+	NK	Laparoscopic removal of cesarean scar pregnancy	45	NK	CS at 30 wk because of abruptio placentae
Taşkin et al, ¹³ 2009	6	24	2/1	1	8	+	NK	TVS guidance, potassium chloride-directed injection	NK	NK	Live male, 1 530 g
Dueñas-García and Young, ¹⁴ 2011	7	34	5/3	3	5	NK	NK	Systemic methotrexate	NK	NK	Unremarkable pregnancy
Wang et al, ⁵ 2010 ^b	8	31	3/1	1	7	+	99 544	Hysteroscopic-directed evacuation and D&C ^b	15	24	CS at 38 wk, live singleton
Present case	9	32	4/2	2	9	+	68 500	Hysteroscopic-directed evacuation and D&C	16	3	CS at 34 wk because of preterm labor

^a β -hCG = β human chorionic gonadotropin; CS = lower-segment cesarean section; D&C = dilatation and curettage; NK = not known; TVS = transvaginal sonography.

^bOur case reported in 2010 was similar to the present case except that the method of conception was in vitro fertilization–embryo transfer.

vaginal sonographic guidance, systemic methotrexate, embryo aspiration, hysteroscopy, and laparoscopic management of CSP have emerged as minimally invasive procedures.^{5,8–14} The pregnant courses of patients who underwent fetal reduction with gestational tissue in situ were not uneventful. All babies were born prematurely because of various causes (severe antepartum hemorrhage, eclampsia, abruptio placentae, or preterm labor). Immediate hemorrhage was even encountered in 2 pregnancies, and bilateral hypogastric artery ligation was necessary to stop the bleeding in 1 woman for preservation of the uterus. In contrast, surgical removal of an HCSP carried an uneventful prenatal course and the pregnant woman delivered a live birth at term (**Table 1**).

Selective reduction of CSPs with KCl under ultrasonographic guidance is the most common type of medical treatment.^{8,10,11,13} The potential teratogenic effect of methotrexate forbids its use in the preservation of intrauterine gestation in HCSPs. Direct embryo aspiration may be an alternative but might affect the intrauterine pregnancy.⁹

Removal of the ectopic mass and repair of the uterine defect by laparoscopy or laparotomy was reported to be a safe technique.^{7,12} A disadvantage of this transabdominal approach was that the healthy uterine tissue was not easy to dissect and identify because of the overlying bladder. We previously reported hysteroscopic management of a first-trimester CSP and an HCSP with preservation of intrauterine gestation and delivery at term.^{5,6} When the patient has no desire to continue the pregnancy, the management of HCSP is similar to that of a single CSP; for preservation of the intrauterine gestation, hysteroscopic manipulation should be reduced as minimally as possible because the intrauterine embryo might be disturbed by the upward force of the distended medium during hysteroscopy. The hysteroscope should not be placed beyond the endocervical canal and should be withdrawn immediately and replaced by placenta forceps and suction curettage to remove the gestational tissues under sonographic guidance after identifying the location of the CSP and coagulating the vessel beds of the implantation site.

CONCLUSION

HCSP is a rare form of ectopic pregnancy, and it mainly occurs after in vitro fertilization–embryo transfer. In our patient this condition occurred spontaneously. The preservation of the normally implanted embryo in HCSP remains more challenging. Although fetal reduction with KCl is frequently adopted, on the basis of the reported cases and our experience, we suggest that hysteroscopic management is another reasonable option but is not necessarily better and requires more data. Ultrasonography can provide a reliable and early diagnosis of HCSP. Early diagnosis is warranted to preserve fertility and avoid maternal morbidity.

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