

Patient's Refusal of Early Treatment of a Cesarean Scar Pregnancy Resulting in Life - Saving Hysterectomy at 23 Weeks Gestation: Report of a Case

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Cesarean scar pregnancy (CSP) is a rare life-threatening form of ectopic pregnancy embedded in the myometrium of a previous cesarean scar. Pathogenesis is suggested to be related with an existing scar defect or a microscopic dehiscence tract generated between the prior cesarean scar and the endometrial canal. The most common risk factor is a history of previous cesarean section. Sonography is the first-line diagnostic tool for the diagnosis of CSP. It is possible to make the diagnosis in the early weeks of pregnancy and early diagnosis would let the patient retain her future fertility. Treatment options prior to rupture include expectant management, dilatation&curettage, conservative medical treatment, hysteroscopic-laparoscopic-primary open surgical removal or hysterectomy. We herein present a case of Cesarean scar ectopic pregnancy complicated with uterine rupture at 23 weeks' gestation.

Key Words: Cesarean scar pregnancy, Ultrasound, Uterine rupture

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Cesarean scar pregnancy (CSP) is a rare life-threatening form of ectopic pregnancy. Its incidence has been increased for the past five years parallel with the increasing rates of cesarean sections as well as increased ability to diagnose the disease.

An ectopic pregnancy embedded in the myometrium of a previous cesarean scar is defined as a cesarean scar ectopic pregnancy.¹ In a CSP, the gestational sac is completely surrounded by the myometrium and the fibrous tissue of a previous cesarean scar separated from the endometrial cavity or fallopian tube.

Pathogenesis is suggested to be related with an existing scar defect or a microscopic dehiscence tract generated between the prior cesarean scar and the endometrial canal.² Such a defect or tract may develop from the trauma of a prior uterine manipulation such as curettage, cesarean delivery, myomectomy, metroplasty, hysteroscopy, and even manual removal of the placenta.³

The most common risk factor associated with CSP is a history of previous cesarean section. But, whether number of pre-

vious cesarean sections might have any role are unclear. Also, time of interval between the previous cesarean section and the subsequent one, indication of the cesarean delivery, placental defects, breech presentation, surgical technique have been discussed as risk factors and need further investigation.⁴⁻⁶ Additional potential risk factors are history of pelvic inflammatory disease or an intrauterine device (IUD).

Sonography is the first-line diagnostic tool for the diagnosis of CSP. By far, it is possible to make the diagnosis in the early weeks of pregnancy and early diagnosis would let the patient retain her future fertility. Strict ultrasound diagnostic criteria have been described in the literature.^{6,7} Additional diagnostic information can also be obtained by magnetic resonance imaging and laparoscopy or hysteroscopy.

When the diagnosis is made prior to rupture, treatment options include expectant management, dilatation & curettage, conservative medical treatment, hysteroscopic-laparoscopic-primary open surgical removal or hysterectomy. A delay in either diagnosis or treatment can lead to catastrophic complications like uterine rupture and massive bleeding necessitating hysterectomy.

We herein present a case of Cesarean scar ectopic pregnancy complicated with uterine rupture at 23 weeks' gestation.

Case Report

A 27-year-old woman with two previous cesarean sections presented at 6 weeks 5 days gestation in her fourth pregnancy with the complaint of lower abdominal pain and vaginal spotting for 12 hours.

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Her obstetric history included a first trimester miscarriage followed by two term lower segment cesarean sections for arrest of labor and previous cesarean section respectively. The last cesarean section was performed 4 years ago. She had no history of any gynecological operation, pelvic inflammatory disease, or intrauterine device (IUD).

On admission to hospital, her vital signs were within the normal range. Her abdominal examination revealed some tenderness in the lower abdomen. Her pelvic examination revealed a slightly enlarged uterus and moderate amount of clots in the vagina. The cervix was uneffaced and was localized in the posterior. The initial blood test revealed a hemoglobin concentration of 12,6 g/dl. A quantitative β -human chorionic gonadotrophin (β -hCG) was >10000 IU/L.

An ultrasound examination by both two-dimensional and three-dimensional ultrasonographic devices (Voluson 730 D Pro, version 4,03; General Electric, Milwaukee, WI, USA) demonstrated a gestational sac bulging into an area of thinned myometrium anteriorly to within 3 mm of the posterior aspect of the bladder, just above the level of the cervix containing an embryo of 8,5 mm crown-rump length (Figures 1,2). Additionally, a prominent peritrophoblastic blood flow has been demonstrated by Doppler flow sonography. The presenting subject fulfilled all of the sonographic criteria described in the literature.^{6,7}

The patient was clearly informed about the high risk life-threatening complications including subsequent uterine rupture and massive bleeding and offered termination of pregnancy (TOP) with the diagnosis of CSP. She refused a TOP and wanted to continue the pregnancy. Although maternal well-being was our first priority, we had to consider the expectant management. As the patient was hemodynamically stable and asymptomatic during hospitalization, a follow-up plan was scheduled and she was discharged on the sixth day.

Eventhough we agreed with her on a weekly follow-up schedule, she was lost to follow-up after 12 weeks of gestation.

At 23 weeks' gestation, she was referred to our department with massive bleeding, hemodynamic compromise with pallor, hypotension, unconsciousness, and a hemoglobin value of 9,9 g/dl, platelet count of $39000/\text{mm}^3$. In fact, she had first admitted another hospital with massive bleeding, laparotomy had been proceeded with an indication of placenta previa and seven units of packed red blood cells had been transfused before admission to our department because of a hemoglobin level of 3 g/dl. After we scanned her records, we proceeded laparotomy under general anaesthesia, via previous Pfannenstiel incision. Intraoperative inspection revealed a significant hemoperitoneum with an estimated 1500 ml of blood and clots and the placenta extruding through the myometrial defect. At

first bilateral uterine artery ligation and then bilateral hypogastric artery ligation were performed, but as bleeding continued we had to perform a subtotal life-saving hysterectomy. At the end of the operation her blood pressure was 60/40 mmHg. Hemoglobin value was 10,0 g/dl, platelet count was $12000/\text{mm}^3$, fibrinogen, antitrombin activity, prothrombin time, partial thromboplastin time, and d-dimer were in normal ranges. Estimated intraoperative blood loss was about 4000 ml. 9 units of whole blood, 6 units of fresh frozen plasma, 2 units of thrombocyte were transfused intraoperatively. She was followed in the antenatal unit for 9 days. She was discharged on the postoperative 22nd day with an uncomplicated recovery.

Histopathology revealed placenta percreata in the wall of the uterus. Other walls were all covered with widespread placenta accreta.



Figure 1: Two dimensional ultrasonographic picture of an ectopic cesarean scar pregnancy implanted within the isthmic area of the lower anterior uterine wall is shown. Endometrial lining, myometrium and gestational sac are demonstrated with thick arrows respectively.



Figure 2: Three dimensional ultrasonographic picture of an ectopic cesarean scar pregnancy implanted within the isthmic area of the lower anterior uterine wall is shown. Endometrial lining, gestational sac and fetal pole are demonstrated with thick arrows respectively.

Discussion

Implantation of a pregnancy within a cesarean scar is a rare form of ectopic pregnancy. Its incidence ranges from 1/1800 to 1/2216 pregnancies.^{4,7} Seow et al have reported the incidence of CSP in women with a previous cesarean section as 0.15%.⁷

Presentation ranges from 5-6 weeks' to 16 weeks' gestation. Usually the presenting symptom is painless vaginal bleeding. Vaginal bleeding may vary from spotting to severe hemorrhage. Low abdominal pain may accompany vaginal bleeding or may be the only sign. Severe abdominal pain with hemodynamic instability and massive hemorrhage may reflect a sign of uterine rupture. Rarely patients may be asymptomatic.

Pathophysiology mainly depends on abnormal implantation of the gestation sac over the scar of a previous cesarean scar. Invasion of the myometrium by the conceptus through a microscopic dehiscence or a scar secondary to poor vascularization of the lower uterine segment with fibrosis and incomplete healing results in abnormal implantation.^{8,9} The mechanism that most probably explains CSP is a history of previous cesarean section. From this point of view, whether the risk of CSP is related to the number of previous cesarean sections has been argued in the literature. While Jurkovic et al⁴ have reported that 72% of the patients with CSP had more than two cesarean deliveries, Seow et al⁷ have reported that 25% of the patients had two prior cesarean sections. Also Rotas et al⁹ have supported that the number of cesarean deliveries has no impact.

Several techniques have been used for the diagnosis of CSP. Ultrasound is the primary diagnostic modality with a sensitivity of 84.6% in the first few weeks of conception.⁹ The following ultrasonographic criteria have been described for the diagnosis of a CSP:^{6,7}

- 1- Empty uterus with clearly demonstrated endometrium
- 2- Empty cervical canal, without a gestational sac or ballooning at the early diagnosis
- 3- On a sagittal view of the uterus, a discontinuity in the anterior uterine wall when running through the amniotic sac is demonstrated
- 4- The gestational sac with or without an embryo presenting fetal cardiac activity is visualized in the anterior localization of the isthmic part of the uterus with a diminished myometrial layer between the bladder and the sac. For the diagnosis of the condition, all the criteria have to be met. Doppler ultrasound, three-dimensional ultrasound, magnetic resonance imaging, hysteroscopy and laparoscopy have been used as adjunct to ultrasound scan. Doppler sonography may demon-

strate a prominent, high velocity-low impedance blood flow surrounding the ectopic pregnancy mass. We suggest to use these modalities in case of uncertainty to confirm the diagnosis.

Once an empty endometrial cavity and a gestational sac in the lower uterine segment are diagnosed on ultrasonographic examination distinction between CSP, cervicoisthmic pregnancy, inevitable miscarriage, and a lowly implanted intrauterine pregnancy should be made. Bleeding is heavier in cervicoisthmic pregnancy and the sonographic diagnosis includes findings that the uterine cavity is empty, gestational sac is located below the internal cervical os, and the cervical canal is distended and barrel-shaped.¹¹ Also, myometrium between the bladder and the gestational sac would be visible in a cervicoisthmic pregnancy. Ultrasound scan of an inevitable miscarriage should reveal a gestational sac be seen in the cervical canal and an avascular appearance on Doppler imaging. However, as the gestation progresses, differentiation between these entities would be difficult. In a lowly implanted intrauterine pregnancy, gestational sac is located inside the endometrial cavity and the myometrium is thick enough to distinguish from a CSP.

Generally, as soon as the diagnosis of a CSP is made in the first trimester, a TOP should be recommended. As the pregnancy continues within the uterus, the risk of placenta accreta is increased up to three- to five-fold. Also, the risk of subsequent uterine rupture and life threatening complications are increased. So an early diagnosis and management should be the aim. Because of the rarity, there are no universal guidelines for management and a variety of therapeutic options have been suggested.⁶⁻¹⁰ Expectant management should be avoided, because the prognosis of an uneventful term pregnancy is poor. Curettage can rupture the uterine scar implantation and lead to severe hemoperitoneum. Systemic and local methotrexate, local potassium chloride, hyperosmolar glucose, and crystalline trichosanthin injections have been performed successfully. Besides minimally invasive interventions such as laparoscopy or hysteroscopy have been suggested, because of the microscopic dehiscence or the scar theory, some believe in the surgical resection of the cesarean scar via open surgery and close the myometrial defect. In the acute setting of uterine rupture, probably hysterectomy is the only option.

In conclusion, early diagnosis and management of CSP would help preserve fertility and avoid life-threatening complications. So, clinicians must consider the possibility of a cesarean scar pregnancy in a woman with a previous uterine surgery.

Sezaryen Skar Gebeliğinde Erken Tedaviyi Reddeden Hastada 23. Gebelik Haftasında

Hayat Kurtarıcı Histerektomi Vakası:

Olgu Sunumu

Sezaryen skar gebeliği, geçirilmiş sezaryen skar hattında myometriuma yerleşmiş hayati tehdit eden nadir bir ektojik gebelik formudur. Patogenezin mevcut bir skar defekti ya da geçirilmiş sezaryen skar ile endometrial kanal arasında oluşmuş mikroskopik dehisans traktusu ile ilişkili olduğu savunulmaktadır. En sık karşılaşılan risk faktörü geçirilmiş sezaryen ökyüsüdür. Sezaryen skar gebeliğinde ilk basamak tanı aracı ultrasonografidir. Erken gebelik haftalarında tanı konulması mümkündür ve erken tanı hastanın fertilitasını korumasına imkan sağlar. Rüptür öncesi tedavi seçenekleri arasında gözlemsel tedavi, dilatasyon&küretaj, konservatif tıbbi tedavi, histeroskopik-laparoskopik-primer açık cerrahi ya da histerektomi sayılabilir. Burada, 23. gebelik haftasında uterus rüptürü ile komplike olmuş bir sezaryen skar ektojik gebelik olgusu sunulmaktadır.

Anahtar Kelimeler: Sezaryen skar gebeliği, Ultrasonografi, Uterin rüptür

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