

Diagnosis and management of cesarean scar pregnancy

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Summary

Objective: To evaluate the diagnosis and management modalities of cesarean scar pregnancy according to our experience. **Design and Setting:** Retrospective study at the Women's Health Research and Education Hospital. **Patients:** Six patients were diagnosed and treated for cesarean scar pregnancy (CSP) with dilatation and curettage, methotrexate (MTX), or laparotomy. **Results:** One patient chose the surgical option due to her desire to have a tubal ligation. In the second case methotrexate was applied initially, but two weeks later suction curettage was applied due to abdominal pain and vaginal bleeding. Suction curettage was used as an initial treatment for four patients. There were not any complications in three of four patients. One patient had heavy vaginal bleeding which started after curettage. On ultrasonographic examination, increasing hemorrhage was seen between the uterus and the bladder so subtotal hysterectomy was performed. **Discussion:** Ultrasound should be used effectively in evaluation of pregnant patients with previous cesarean deliveries. There is still no unique treatment modality for CSP, so treatment should be tailored for each patient. Before the 7th week, abortion should be considered. After the 7th week, MTX and/or surgical options should be preferred.

Key words: Scar pregnancy; Ectopic pregnancy; Management; Ultrasound.

Introduction

Cesarean scar pregnancy (CSP), a very rare form of ectopic pregnancy, is located outside the uterine cavity and completely surrounded by myometrium in the prior lower uterine segment [1]. Because it often is misdiagnosed as cervical or aborting pregnancy, CSP may result in life treating conditions such as uterine rupture, hemorrhage, disseminated intravascular coagulation and even maternal death [1-3]. There is no accepted management protocol, so because of the rarity of this life-threatening disease, each patient should be evaluated individually [1].

The aim of this study was to evaluate the diagnosis and management modalities of CSP at our clinic.

CSP is more common than previously thought. It seems to be on the rise because of increased cesarean delivery rates as well an improvement in the clinical knowledge of clinicians and high clinical index of suspicion [2]. The estimated incidence of CSP ranges from 1:1,800 to 1:2,216 pregnancies [5, 6], but the true incidence has not been determined because so few cases have been reported in the literature [7].

The diagnosis of CSP is mainly accomplished by combining transvaginal sonography (TVS) with Doppler flow imaging [5, 7]. Once the correct diagnosis is made CSP should be terminated to avoid life-threatening complications [7, 8].

Materials and Methods

We searched and analyzed the medical records of patients admitted to our department with a diagnosis of cesarean scar pregnancy from January 2005 to March 2011. There were 12

hospital admissions with the diagnosis of CSP, but six of 12 patients had abortions when re-examined carefully later on.

The diagnosis of CSP was determined based on the following ultrasonographic criteria: 1) empty uterus; 2) empty cervical canal; 3) anteriorly located gestational sac with a diminished myometrium layer between the bladder and the sac; 4) discontinuity in the anterior uterine wall of the uterus on a sagittal view of the uterus after the gestational sac (Figure 1).

Ethical committee approval was given for the study.

Results

The mean maternal age was 32 (range 28-38 years). Five patients had undergone two previous cesarean sections and one patient had undergone four previous cesarean sections. The mean gestational age during the diagnosis was six weeks and two days (range 4 weeks and 4 days - 12 weeks). Fetal cardiac activity was present in only two

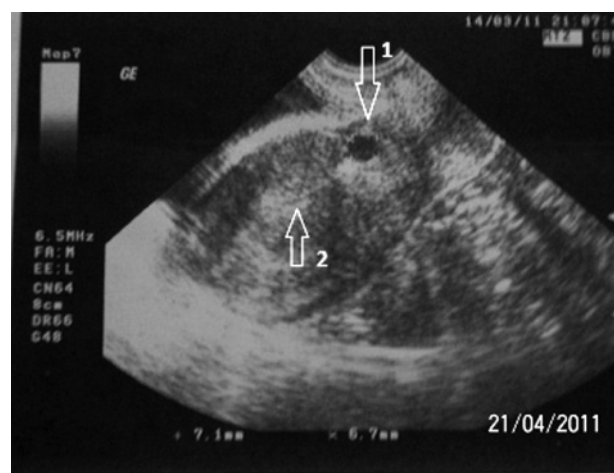


Figure 1. — Gestational sac in scar tissue (arrow 1); Empty appearance of uterine cavity (arrow 2).

Revised manuscript accepted for publication September 26, 2011

Table 1. — *Characterisations and treatment of cases*

Case	Age	Presenting symptoms	Number of previous cesarean sections	GA (week)	Fetal viability	Initial management	Complications	Additional management
1	33	-	4	CRL = 10.7 mm 7 w / 4 d	+	Laparotomy/ hysterotomy	Bladder injury	-
2	38	-	2	CRL = 9.9 mm 7w	-	Systemic 1 mg/kg IM methotrexate	-	Curettage
3	28	Slight Vaginal bleeding Pelvic pain	2	GS = 17.6 mm 6 w	-	Curettage	-	-
4	30	Pelvic pain	2	CRL = 47.7 mm 12 w	-	Curettage	Severe vaginal bleeding	Laparotomy/ hysterectomy
5	32	Slight Vaginal bleeding	2	CRL = 10.2 mm 7 w	+	Curettage	-	-
6	32	Pelvic pain	2	GS = 6 mm 4 w / 4 d	-	Curettage	-	-

CRL: crown rump length, GS: gestational sac.

of the pregnancies. The characteristics of the six patients are given in Table 1. Average gestational age of the patients was six weeks and two days. Fetal pulse of two of those had been detected with ultrasound. In the other four cases, fetal pulse had not been detected.

Clinical presentations were described as follows: mild vaginal bleeding and abdominal bleeding (one patients), mild abdominal pain (two patients) and vaginal bleeding (one patient) and asymptomatic (two patients). Asymptomatic patients were diagnosed during the routine first trimester ultrasonographic (US) screening.

After explaining all the treatment modalities, one patient chose the surgical option due to desire of tubal ligation. The gestational mass was excised and the uterine segment was repaired. During the operation the bladder was damaged due to adhesions and the defect was repaired with success. In the second case methotrexate (1 mg/kg IM, single dosage) was applied initially, but two weeks later suction curettage was applied due to abdominal pain and vaginal bleeding. There were no complications during or at the end of the curettage. Suction curettage was used as an initial treatment for four patients. There were not any complications in three of four patients. One patient had heavy vaginal bleeding which started after curettage. On US examination, increasing hemorrhage was seen between the uterus and the bladder and subtotal hysterectomy was performed.

The results of the study were sent as an abstract to ASRM 2011.

Discussion

CSP is rare and often misdiagnosed as other diseases like miscarriage and cervico-isthmic pregnancy [3, 8]. What is noteworthy in our research is that before 2010 there had been

no admissions to our hospital with this diagnosis. Moreover, after this date, the six patients who were prediagnosed with CSP had been thoroughly examined and checked by a US device with a strong resolution and either miscarriages or missed abortions had been detected. This situation clearly shows we have information about CSP due to recent case reports and we acted in an over-sensitive manner every time we encountered a pregnant patient with a cesarean history.

Although CSP is rare, without a high index of suspicion and early diagnosis, this abnormal implantation can lead to uterine rupture, hemorrhage, serious maternal morbidity and loss of future fertility, and even maternal death [7, 9].

The invasion of the myometrium through a microscopic tract is the most probable mechanism of development of CSP. This microscopic tract may have occurred from trauma of previous uterine surgeries like cesarean section, myomectomy, dilatation and curettage (D&C), and manual removal of the placenta. However, the exact cause of CSP is unknown [1, 5, 9, 10].

CSP is more common than previously thought and it seems to be on the rise because of increase in cesarean delivery rates as well as improvement in the clinical knowledge of clinicians and high clinical index of suspicion [4]. The estimated incidence of CSP ranges from 1:1,800 to 1:2,216 pregnancies [5, 6], but the true incidence has not been determined because so few cases have been reported in the literature [7]. What attracted our attention is until the year 2010, there had not been any hospital admissions with the diagnosis of CSP. In contrast, after the year 2010, it was noted that six of 12 hospital admissions with the diagnosis of CSP were abortion or miscarriage cases after carefully being re-examined. This indicates that increased knowledge of CSP due to reported case reports led us to over-diagnose and be oversuspicious about pregnancies with prior cesarean deliveries.

Recently there has been an increase in reported CSP cases. Donald *et al.* [11] reported 19 CSP cases between 1966 and 2002 in a literature review study whereas Homayoun *et al.* reported 268 cases between 1995 and 2008 [4]. In our study, we diagnosed and treated six cases of CSP in a 3-month period. The recent increase in the number of cases may reflect high cesarean birth rate worldwide, but it also may be associated with better diagnostic accuracy, improved knowledge, and high index of suspicion [1, 12]. The association between cesarean deliveries for breech presentation and subsequent CSP is reported in the literature. They hypothesis is that most indications for prior cesarean deliveries in CSP cases are elective, and thus there is a poorly developed lower uterine segment that leads to incomplete healing [9, 13, 14]. Some authors have proposed that the increase of these abnormal pregnancies may be due to change in surgical technique of the hysterotomy. In the past double-layer closure was performed, with sutures inverting the first layer by the second one. As commonly used today, closure of hysterotomy with monolayer noninverting running sutures leads to incomplete postoperative healing and creation of defects in scar tissue [1, 4, 13].

Chuang *et al.* reported no association between number of cesarean sections and CSP occurrence [15], but Jurkovic *et al.* reported that the number of cesarean sections affected the occurrence of CSP because the scar surface is increased and the anterior uterine wall may be deficient because of poor vascularity, fibrosis and incomplete healing [6]. In the present study, five of six patients had undergone two previous cesarean sections and one patient had undergone four previous cesarean sections which is compatible with Jurkovic *et al.*'s study.

There are very different presenting manifestations in CSP. It may be present with painless vaginal bleeding, lower abdominal pain, vaginal bleeding plus abdominal pain, and it may be an incidental finding on routine ultrasonography in an asymptomatic woman [1, 3, 4]. In the present study, the patients' clinical presentations also ranged from asymptomatic to mild abdominal pain and vaginal bleeding.

There is no consensus on the best method and criteria to diagnose cases. However the majority of CSPs have been confirmed by TVS. Maymon *et al.*, recommend using combined TVS and transabdominal sonography (TAS) with a full bladder. Thus a 'panoramic view' of the uterus is provided with accurate measurement of the distance between the gestational sac and bladder [13]. Occasionally, additional diagnostic modalities such as Doppler flow imaging, three-dimensional ultrasonography, magnetic resonance imaging, and even invasive procedures such as hysteroscopy and cystoscopy may be necessary for the differential diagnosis between cervical pregnancy, cervico-isthmic pregnancy, spontaneous miscarriage in progress and CSP. True diagnosis is crucial because a large number of complications caused by misdiagnosis leads to expectant management and inappropriate interventions [4, 13, 16, 17].

Our all patients were diagnosed by a combination of TVS and TAS. The diagnosis of cesarean scar pregnancy

was determined based on the following ultrasonographic criteria: 1) empty uterus; 2) an empty cervical canal; 3) anteriorly located gestational sac with a diminished myometrium layer between the bladder and the sac, and 4) discontinuity in the anterior uterine wall of the uterus on a sagittal view of the uterus following the gestational sac [11, 14, 18].

Because of the infrequency of CSP, there are no universal treatment guidelines for this abnormal pregnancy, although several treatment modalities have been recommended. All reports consist of few cases and there is no agreement on which treatment modality should be preferred [2, 4]. In cases when the patient might request the continuation of the pregnancy, the patient should be thoroughly informed about the possible complications and if the pregnancy is continued it must be closely monitored. Due to the high risk of uterine rupture, invasive placenta and profuse uterine bleeding, the current trend is termination of CSP after the explanation of risks to couples [13, 16].

In a hemodynamically stable patient two management options may be considered: medical or surgical intervention. Surgical intervention has been successfully performed either laparoscopically [19] or by laparotomy [14, 17] in the form of excision of the ectopic pregnancy and repair of the myometrium. However, the surgical operation carries a significant risk of uncontrolled hemorrhage, resulting in hysterectomy and loss of future fertility in some of cases [6, 20].

In one of our four cases, laparotomy was used as an early treatment. The reason for this was the request for tubal ligation and the preference of surgical treatment by the patient. Excision of the scar pregnancy and treatment of the scar was achieved but, during surgery the bladder had been damaged and had to be treated.

Evacuation of CSP by curettage alone has been performed, but secondary salvage treatment has already been proven necessary. Therefore, D&C should not be considered as the first choice of therapy. This is because the majority of the villi are implanted in the myometrium and it seems unlikely that the gestational sac can be expelled by curettage without perforating the uterine wall or damaging the urinary bladder, and may also cause life-threatening hemorrhage [11, 13]. In contrast, some authors have reported successfully treating CSP by D&C under the guidance of ultrasound, without complications [6-7, 21].

Suction curettage was used as the initial treatment on four of our patients. We could not successfully manage in only one of these patients because the patient's gestational weeks were more than 12 weeks. The other three successfully treated pregnancies were less than seven weeks of gestation. The gestational age is an important factor when curettage is the treatment of choice.

Medical therapy is noninvasive and avoids further damage to the uterus, and therefore may maximize the chance of preservation of the uterus in patients who desire fertility. However, the efficacy and safety of this treatment modality is still unknown [8].

Medical treatment consists of methotrexate (MTX) administered either systemically [22, 23], locally [4] or

combined [5, 15]. Because CSP is surrounded by fibrous scarring rather than normally vascularized myometrium, potentially limiting systemic access, injecting MXT directly into the gestational sac may be more effective [11]. Due to the rarity of CSP, it is impossible to conclude whether systemic or direct intrascar administration of MTX is safer or more effective. Patients receiving primary medical treatment of a scar ectopic pregnancy should be monitored closely, preferably in a hospital [8]. Medical treatment should be considered in women with no or slight symptoms and stable vital signs, simultaneously with sonography showing no signs of uterine rupture [24]. In one of our cases MTX (1 mg/kg IM, single dosage) was applied initially, but after two weeks suction curettage was applied due to abdominal pain and vaginal bleeding. There were no complications during or at the end of the curettage.

Methotrexate therapy may be combined either with an intrascar potassium chloride injection as an embryocide, with TVS guided aspiration of the scar pregnancy [9, 25] or with uterine artery embolization to minimize hemorrhage [11, 26]. Some authors, like Jiao *et al.*, proposed that selective uterine artery embolization can temporarily block uterine perfusion and minimize hemorrhage and they have used this technique before or after curettage [24].

Although Maymon *et al.* reported that no single management modality is entirely reliable and that none can guarantee uterine integrity [13], Fylstra proposed that primary surgical treatment by laparotomy and hysterotomy may be the best treatment option, because only surgical resection offers the opportunity to remove the pregnancy and simultaneously repair the defect [11].

In conclusion, due to the upward trend in cesarean sections, clinicians should always consider CSP in the differential diagnosis. During this evaluation, especially the US evaluation in the first trimester, the observation of an empty uterine cavity is also important.

In diagnosing CSP, TAS and TVS should be used in combination to obtain an accurate diagnosis. Every clinician should have knowledge of different treatment modalities about CSP and the treatment should be tailored to each patient. In patients with gestational age less than seven weeks, curettage can be performed, but in greater gestational age curettage should not be preferred, and medical or surgical treatment modalities should be chosen as the treatment modality.

References

- [1] Rotas M.A., Haberman S., Levгур M.: "Cesarean scar ectopic pregnancies: etiology, diagnosis and management". *Obstet. Gynecol.*, 2006, 107, 1373.
- [2] Einkenkel J., Stump P., Kosling S., Horn L.C., Hockel M.: "A misdiagnosed case of cesarean scar pregnancy". *Arch. Gynecol. Obstet.*, 2005, 271, 178.
- [3] Hasewaga J., Ichizuka K., Matsuoaka R., Otsuki K., Sekizawa A., Okai T.: "Limitations of conservative treatment for repeat cesarean scar pregnancy". *Ultrasound Obstet. Gynecol.*, 2005, 25, 310.
- [4] Sadeghi H., Rutherford T., Rackow B.W., Campbell K.H., Duzyj C.M., Guess M.K. *et al.*: "Cesarean scar ectopic pregnancy: case series and review of the literature". *Am. J. Perinatol.*, 2010, 27, 111.
- [5] Seow K.M., Huang L.W., Lin Y.H., Lin M.Y., Tsai Y.L., Hwang J.L.: "Cesarean scar pregnancy: issues in management". *Ultrasound Obstet. Gynecol.*, 2004, 23, 247.
- [6] Jurkovic D., Hillaby K., Woelfer B., Lawrence A., Salim R., Elson C.J.: "First trimester diagnosis and management of pregnancies implanted into the lower uterine segment cesarean section scar". *Ultrasound Obstet. Gynecol.*, 2003, 21, 220.
- [7] Maymon R., Halperin R., Mendlovic S., Schneider D., Vaknin Z., Herman E., Pansky M.: "Ectopic pregnancies in caesarean section scars: the 8 year experience of one medical centre". *Hum. Reprod.*, 2004, 19, 278.
- [8] Lam P.M., Lo K.W., Lau T.K.: "Unsuccessful medical treatment of cesarean scar ectopic pregnancy with systemic methotrexate: a report of two cases". *Acta Obstet. Gynecol. Scand.*, 2004, 83, 108.
- [9] Tan G., Chong Y.S., Biswas A.: "Caesarean scar pregnancy: a diagnosis to consider carefully in patients with risk factors". *Ann. Acad. Med. Singapore*, 2005, 34, 216.
- [10] Neiger R., Weldon K., Means N.: "Intramural pregnancy in a cesarean section scar. A case report". *J. Reprod. Med.*, 1998, 43, 999.
- [11] Fylstra D.L.: "Ectopic pregnancy within a cesarean scar: a review". *Obstet. Gynecol. Surv.*, 2002, 57, 537.
- [12] Leitch C.R., Walker J.J.: "The rise in cesarean section rate: the same indications but a lower threshold". *Obstet. Gynecol. Surv.*, 1999, 54, 19.
- [13] Maymon R., Halperin R., Mendlovic S., Schneider D., Herman A.: "Ectopic pregnancies in a caesarean scar: review of the medical approach to an iatrogenic complication". *Hum. Reprod. Update*, 2004, 10, 515.
- [14] Vial Y., Petignat P., Hohlfield P.: "Pregnancy in a cesarean scar". *Ultrasound Obstet. Gynecol.*, 2000, 16, 592.
- [15] Chuang J., Seow K.M., Cheng W.C., Tsai Y.L., Hwang J.L.: "Conservative treatment of ectopic pregnancy in a cesarean section scar". *BJOG*, 2003, 110, 869.
- [16] Wang C.B., Tseng C.J.: "Primary evacuation therapy for cesarean scar pregnancy: three new cases and review". *Ultrasound Obstet. Gynecol.*, 2006, 27, 222.
- [17] Valley M.T., Pierce J.G., Daniel T.B., Kaunitz A.M.: "Cesarean scar pregnancy: imaging and treatment with conservative surgery". *Obstet. Gynecol.*, 1998, 91, 838.
- [18] Godin P.A., Bassil S., Donnez J.: "An ectopic pregnancy developing in a previous caesarian section scar". *Fertil. Steril.*, 1997, 67, 398.
- [19] Wang Y.L., Su T.H., Chen H.S.: "Laparoscopic management of an ectopic pregnancy in a lower segment cesarean section scar: a review and case report". *J. Minim. Invasive Gynecol.*, 2005, 73.
- [20] Graesslin O., Dedeker F. JR, Quereux C., Gabriel R.: "Conservative treatment of ectopic pregnancy in a cesarean scar". *Obstet. Gynecol.*, 2005, 105, 869.
- [21] Arslan M., Pata O., Dilek T.U.K., Aktas A., Aban M., Dilek S.: "Treatment of viable cesarean scar ectopic pregnancy with suction curettage". *Int. J. Gynecol. Obstet.*, 2005, 89, 163.
- [22] Shufaro Y., Nadjari M.: "Implantation of a gestational sac in a cesarean section scar". *Fertil. Steril.*, 2001, 75, 1217.
- [23] Ravhon A., Ben-Chetrit A., Rabinowitz R., Neuman M., Beller U.: "Successful methotrxate treatment of a viable pregnancy within a thin uterine scar". *Br. J. Obstet. Gynecol.*, 1997, 104, 628.
- [24] Jiao L.Z., Zhao J., Wan X.R., Liu X.Y., Feng F.Z., Reng T., Xiang Y.: "Diagnosis and treatment of cesarean scar pregnancy". *Chin. Med. Sci. J.*, 2008, 23, 10.
- [25] Hwu Y.M., Hsu C.Y., Yang H.Y.: "Conservative treatment of cesarean scar pregnancy with transvaginal needle aspiration of the embryo". *BJOG*, 2005, 112, 841.
- [26] Ghezzi F., Lagana D., Franchi M., Fugazzola C., Bolis P.: "Conservative treatment by chemotherapy and uterine arteries embolization of a cesarean scar pregnancy". *Eur. J. Obstet. Gynecol. Reprod. Biol.*, 2002, 103, 88.

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