

Conservative treatment by endoscopy of a cesarean scar pregnancy: two case reports

C. Colomé, M.D.; M.T. Cusidó, M.D., PhD.; L. Hereter, M.D.; M.A. Pascual, M.D., PhD.;
R. Fábregas, M.D.

Department of Obstetrics, Gynecology and Human Reproduction, Institut Universitari Dexeus, Barcelona (Spain)

Summary

Background: Cesarean section scar pregnancy is the rarest form of ectopic pregnancy and the most dangerous due to the high risk of uterine rupture and hemorrhage. **Case:** We present two case reports of women diagnosed with an ectopic cesarean scar pregnancy. We performed conservative treatment because both patients desired fertility preservation. The first case was treated with laparoscopy and hysteroscopy simultaneously. For the second case the treatment started with an ultrasound-guided injection of methotrexate. Surgical laparoscopy and hysteroscopy were subsequently performed simultaneously. Four months later, the first woman had a spontaneous singleton pregnancy. An elective cesarean was performed. **Conclusion:** In these two case reports we have presented our experience with endoscopic surgery in the management of two patients who had a cesarean scar pregnancy and desired to preserve their fertility.

Key words: Cesarean scar pregnancy; Laparoscopic surgery; Hysteroscopy surgery; Methotrexate; Transvaginal ultrasound.

Introduction

Cesarean section scar pregnancy is a form of ectopic pregnancy, with a high risk of uterine rupture and hemorrhage, hence the need for termination [1, 2]. It was first described in 1978 by Larsen and Salomón [3], and only a further 19 cases were published in the period up to 2001. However, over the last six years there has been a notable rise in reports of cesarean scar pregnancy (CSP) in the English language literature. Ash *et al.* [4] pointed out that this must be secondary to an increase in the number of cesareans being performed, as well as to improved diagnosis of the condition.

Although the incidence of CSP is actually low (the estimate being 1: 2,226 of all pregnancies), the large rise in the number of cesareans now performed would suggest a concomitant increase in this type of pregnancy [5]. However, the exact incidence of this type of pregnancy is difficult to determine and estimates vary widely: Seow *et al.* [5] reports a rate of 6.1% in patients with a history of ectopic pregnancy and cesarean scar (at least one) or hysterotomy scar, while Wang *et al.* [6] gives a figure of 21.6%.

The etiology of the condition remains unknown, although some hypotheses refer to trophoblast invasion of the myometrium when there is a history of cesarean section (rising by up to 60-70% in the case of multiple cesareans, dilation and curettage or adenomyosis) [2].

The symptoms shown by patients can vary widely and although there is often acute or moderate pain Rotas *et al.* [7] reports a diagnostic rate of 37% in asymptomatic patients under routine ultrasound examination.

The development and use of transvaginal ultrasound (TVS) in the first trimester has aided the diagnosis of this

type of pregnancy, and this raises the possibility of conservative treatment [4]. Therapeutic strategies include local (8) or systemic injection of methotrexate (MTX), ultrasound-guided curettage, laparoscopic or laparotomic excision, and hysterectomy, although there is no standard protocol for the diagnosis and treatment [6, 9].

Case Reports

Case 1

The patient was a 38-year-old woman with no history of note except for two previous term cesareans as a result of fetopelvic disproportion and a left salpingo-oophorectomy due to ectopic pregnancy.

She attended our clinic expressing the desire to have a child with her new partner. One month later the patient returned to our clinic complaining of intermittent blood loss and pain in the right iliac fossa. Urine β -hCG was positive and a subsequent TVS revealed an anteverted uterus of 98 x 43 mm with cesarean scar dehiscence (serous) and the presence of a gestational sac with a vitelline vesicle invaginating the scar; both adnexa appeared to be normal with no free fluid in the pouch of Douglas (Figure 1).

Given the suspicion of an ectopic CSP the patient was admitted for treatment. Due to her desire to become pregnant we opted to perform conservative laparoscopic surgery. After opening the cavity we performed uterine scar resection and metroplasty, while simultaneously carrying out hysteroscopic resection of the gestational sac and decidua in the area of the cesarean scar. At the end of the surgery we ensured that the cavity was properly sealed.

After 24 hours the patient was clinically stable and all tests were normal, and she was thus discharged. Follow-up a week later showed β -hCG to be 283 IU/l, and subsequent weekly tests gave values of 26.82 IU/l and 5.48 IU/l, respectively. Analysis of pathological anatomy revealed ovular-decidual remains plus connective tissue with scarce adipose tissue islets and evidence of fibrosis consistent with scar material.

Revised manuscript accepted for publication July 28, 2008

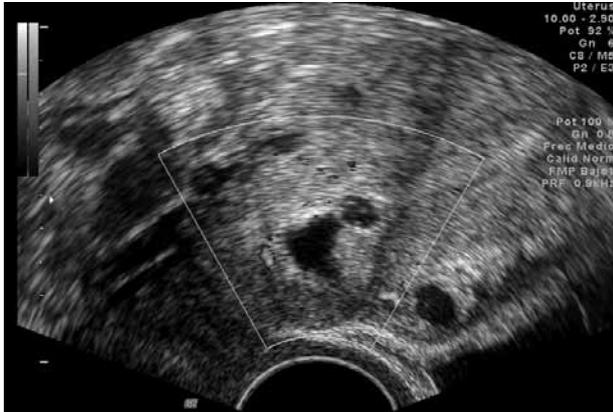


Figure 1. — TVS: cesarean scar dehiscence (serous) with a gestational sac.

Four months later the patient once again attended our clinic due to a spontaneous singleton pregnancy. The pregnancy proceeded without incident and only standard obstetric monitoring was required. An elective cesarean was performed at 37 weeks and a male fetus weighing 3,150 g was delivered. During the cesarean the bladder was observed to be firmly adhered to the uterus, and there was also dehiscence of a previous scar, which was sutured without incident. The patient was discharged five days later and her postoperative evolution was problem-free.

Case 2

The patient was a 36-year-old woman with an unremarkable history except for a previous term cesarean due to fetopelvic disproportion.

She attended our clinic expressing the desire to become pregnant. The case was assessed as infertility secondary to low follicular reserve and male factor, and we thus recommended cycles of IVF-ICSI. Following the first cycle of in vitro fertilization two embryos were transferred on subsequent tests of β -hCG which showed levels of 80.7 and 2,105 five days later. One week later she presented at the emergency department due to heavy blood loss and pain. TVS revealed a retroverted uterus measuring 86 x 59 mm and a blood-filled endometrial cavity. In the area of the previous cesarean scar the echography showed a gestational sac with an embryo and positive fetal heartbeat (Figure 2). The suspected diagnosis was therefore an ectopic



Figure 2. — Echography 3D: gestational sac with an embryo in the area of the previous cesarean scar.

CSP and the patient was admitted for treatment. This began with an ultrasound-guided intrasac injection of 50 mg of MTX to reduce the trophoblastic tissue, the idea being to perform – given the patient's desire to become pregnant – conservative endoscopic surgery 48 hours after medical treatment. Surgical laparoscopy and hysteroscopy were subsequently performed simultaneously. During the laparoscopy we observed bulging of the cesarean scar area and after opening the cavity metroplasty was performed; the opening was then sutured with individual stitches. Hysteroscopy revealed severe hematometra plus dehiscence of the previous cesarean scar, with the abundant presence of necrotized ovular decidual tissue. The abortive remains were then resected and removed via suction curettage. At the end of the intervention we ensured that the cavity was properly sealed.

After 24 hours the patient was clinically stable and all tests were normal, and she was thus discharged. Follow-up a week later showed β -hCG to be 94 IU/l. The analysis of pathological anatomy revealed ovular-decidual remains along with focal adenomyosis.

Discussion

To be able to offer conservative treatment it is necessary to have an early diagnosis, and in the case of ectopic pregnancy this has become possible through the introduction of TVS. The diagnostic ultrasound image shows an empty uterus and cervical canal with a gestational sac in the anterior part of the isthmic portion and reduction of the myometrial wall between the bladder and the gestational sac; this aids differential diagnosis with respect to a cervicoisthmic pregnancy. Cervical ectopic pregnancy is characterized by an empty uterus, a barrel-shaped cervix, a gestational sac present below the level of the uterine arteries, absence of the sliding organ sign and blood flow around the gestational sac on color Doppler. Both Doppler and 3D-ultrasound provide further information for diagnosis and subsequent monitoring. Three-dimensional ultrasonography and its applications allow better images to be obtained thus improving the ability to identify anatomic details that permit a more accurate diagnosis [10, 11].

The differential diagnosis should be made not only with respect to a cervicoisthmic pregnancy but also as regards an incomplete miscarriage that shifts under cervical pressure. The literature does include one case report of a CSP in which an expectant management approach was maintained up to 36 weeks, with the subsequent delivery of a live male fetus [14]. In most cases the evolution of an ectopic cesarean scar pregnancy leads to uterine rupture and consequently, profuse hemorrhaging, thus a termination is required.

Due to the low incidence of this type of pregnancy there are no standard treatment protocols, although proposals for consensus on the diagnosis and treatment of ectopic pregnancies are increasingly to be found in the literature [15].

The approach to this pregnancy can be divided into two broad categories: radical and conservative. Radical hysterectomy is not a therapeutic strategy and is applied only in the case of intractable bleeding and after all conservative methods have failed. Conservative treatment must be

evaluated according to each individual case and the experience of the medical team, the sole objective being to finalize the gestation. Some authors such as Jurkovic *et al.* [16] argue that 44% of pregnancies of this type result in miscarriage. Obviously, the choice of treatment does not depend solely on the patient's desire to have children but also on other factors such as symptomatology, gestational age, the viability of the pregnancy, uteroplacental neovascularization and the patient's preferences.

As regards conservative treatments these can be divided into two types: surgical and medical. Non-surgical treatments offer a range of options, the greatest advantage being that there is no scar resection. Noteworthy among these approaches is conservative systemic treatment with methotrexate, which offers a quick resolution albeit with greater side-effects and the possibility of continued symptoms and the subsequent need to perform surgical laparoscopy [17]. Ultrasound-guided MTX treatment increases concentration in the target area and is associated with fewer side effects. In the series reported by Seow *et al.* [10], 100% of patients were treated successfully, despite the persistence in some cases of post-treatment masses. Another conservative treatment is arterial embolization [18], and some authors combine the two techniques, performing embolization of both uterine arteries and administering an intramuscular injection of MTX [19]. Finally, a little over ten years ago Godin *et al.* [2] reported a case treated successfully with KCl and MTX, while more recently Wang and colleagues [20] have described a case of heterotopic pregnancy in which selective fetocide of the CSP was achieved through administration of KCl.

Surgical treatments such as laparotomy are used when the pregnancy has to be localized, is at an advanced stage, or in the event of significant bleeding. However, the lengthy hospital stay and recovery period required following this type of intervention must be taken into account. Laparoscopic treatment can help to localize those gestations that are deeply implanted in the scar and aid not only diagnosis but also metroplasty and repair of the area, as in the cases described here. When there is doubt as to which type of endoscopy to use Wang *et al.* [6] recommend that the decision is made according to the location of the ectopic pregnancy and the medical team's own experience. With respect to pregnancies that are deeply implanted in a cesarean scar, opinion varies: some groups recommend laparoscopy while others prefer hysteroscopic visualization of the uterine cavity or ultrasound-guided curettage [21].

In both cases reported here we opted to combine laparoscopy with hysteroscopy as we believe this aids surgery and makes it easier to assess whether the cavity is properly sealed once the ectopic pregnancy has been removed. As regards the use of suction curettage its efficacy among these patients remains controversial and most groups complement it with other techniques. Finally, and as reported by the group of El-Matary *et al.* [14], it is possible to take an expectant management

approach to this type of pregnancy provided that the risks involved are constantly assessed.

Although there is no standard protocol for the follow-up of these patients there is a good degree of consensus regarding the need to monitor blood β -hCG and perform periodic TVS examinations.

In terms of recommendations for future pregnancies Seow *et al.* [8] point out that scar repair does not guarantee the safety of subsequent gestations, and note that reports have described an increased incidence of placenta accreta or percreta following such pregnancies. They also recommend the use of elective cesarean plus blood reserve in any subsequent pregnancies, which could be diagnosed by echography. If placenta accreta or percreta is expected special preparations for surgery can be made. This group also recommend that pregnancy be avoided for between three months and up to 1-2 years [5]. At all events any later pregnancies should be accompanied by detailed ultrasound examinations.

Our experience demonstrates the success of conservative treatment to obtain a term pregnancy. However, in all cases we recommend an elective cesarean due to the risk of scar dehiscence.

Finally, given the increased number of cesarean deliveries today clinicians should be alert to the possibility of encountering a cesarean scar pregnancy.

Acknowledgement

The authors are grateful to the "Càtedra d'Investigació en Obstetrícia i Ginecologia" (Universitat Autònoma de Barcelona) for its support.

References

- [1] Arslan M., Pata O., Dilek T.U., Aktas A., Aban M., Dilek S.: "Treatment of viable caesarean scar ectopic pregnancy with suction curettage". *Int. J. Gynaecol. Obstet.*, 2005, 89, 163.
- [2] Godin P.A., Bassil S., Donnez J.: "An ectopic pregnancy developing in a previous caesarean section scar". *Fertil. Steril.*, 1997, 67, 398.
- [3] Larsen J.V., Solomon M.H.: "Pregnancy in a uterine scar sacculus: an unusual cause of postabortal haemorrhage". *Sou. Afr. Med.*, 1978, 53, 142.
- [4] Ash A., Smith A., Maxwell D.: "Caesarean scar pregnancy". *BJOG*, 2007, 114, 253.
- [5] Seow K.M., Huang L.W., Lin Y.H., Lin M.Y., Tsai Y.L., Hwang J.L.: "Caesarean scar pregnancy: issues in management". *Ultrasound Obstet. Gynecol.*, 2004, 23, 247.
- [6] Wang C.J., Chao A.S., Yuen L.T., Wang C.W., Soon Y.K., Lee C.L.: "Endoscopic management of caesarean scar pregnancy". *Fertil. Steril.*, 2006, 85, 494.
- [7] Rotas M.A., Haberman S., Levгур M.: "Caesarean scar ectopic pregnancies: etiology, diagnosis and management". *Obstet. Gynecol.*, 2006, 107, 1373.
- [8] Pascual M.A., Ruiz J., Tresserra F., Sanuy C., Grases P.J., Tur R. *et al.*: "Cervical ectopic twin pregnancy: diagnosis and conservative treatment". *Hum. Reprod.*, 2001, 1, 584.
- [9] Lee C.L., Wang C.J., Chao A., Yen C.F., Soong Y.K.: "Laparoscopic management of an ectopic pregnancy in a previous caesarean section". *Hum. Reprod.*, 1999, 14, 1234.
- [10] Seow K.M., Hwang J.L., Tsai Y.L., Huang L.W., Lin Y.H., Hsieh B.C.: "Subsequent pregnancy outcome after conservative treatment of a previous caesarean scar pregnancy". *Acta Obstet. Gynecol. Scand.*, 2004, 83, 1167.

- [11] Pascual M.A., Hereter L., Graupera B., Tresserra F., Fernandez-Cid M., Simon M.: "Three-dimensional power Doppler ultrasound diagnosis and conservative treatment of ectopic pregnancy in a caesarean section scar". *Fertil. Steril.*, 2007, 88.
- [12] Jurkovic D., Hacket E., Campbell S.: "Diagnosis and treatment of early cervical pregnancy: a review and a report of two cases treated conservatively". *Ultrasound Obstet. Gynecol.*, 1996, 8, 373.
- [13] Timor-Tristch I.E., Monteagudo A., Mandeville E.O., Peisner D.B., Anaya G.O., Pirrone E.C.: "Successful management of viable cervical pregnancy by local injection of methotrexate guided by transvaginal ultrasonography". *Am. J. Obstet. Gynecol.*, 1994, 179, 737.
- [14] El-Matary A., Akinlade R., Jolaoso A.: "Caesarean scar pregnancy with expectant management to full term". *J. Obstet. Gynaecol.*, 27, 6, 624.
- [15] Condous G., Timmerman D., Goldstein S., Valentin L., Jurkovic D., Bourne T.: "Pregnancies of unknown location". *Ultrasound Obstet. Gynecol.*, 2006, 28, 121.
- [16] 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 caesarean section scar". *Ultrasound Obstet. Gynecol.*, 2003, 21, 220.
- [17] Deb S., Clewes J., Hewer C., Raine-Fenning N.: "The management of cesarean scar ectopic pregnancy following treatment with methotrexate: a clinical challenge". *Ultrasound Obstet. Gynecol.*, 2007.
- [18] Yang M.J., Jeng M.H.: "Combination of transarterial embolization of uterine arteries and conservative surgical treatment for pregnancy in a caesarean section scar. A report of 3 cases". *J. Reprod. Med.*, 2003, 48, 213.
- [19] Erin L., Hois J.F. Hibbeln: "Ectopic pregnancy in a caesarean section scar treated with intramuscular methotrexate and bilateral uterine artery embolization". 2007 (www.interscience.wiley.com)
- [20] Wang C.N., Chen C.K., Wang H.S., Chiueh H.Y., Soong Y.K.: "Successful management of heterotopic caesarean scar pregnancy combined with intrauterine pregnancy after in vitro fertilization-embryo transfer". *Fertil. Steril.*, 2007, 88.
- [21] Ben Nagi J., Ofili-Yebovi D., Sawyer E., Aplin J., Jurkovic D.: "Successful treatment of a recurrent Cesarean scar ectopic pregnancy by surgical repair of the uterine defect". *Ultrasound Obstet. Gynecol.*, 2006, 28, 855.

Address reprint requests to:
C. COLOMÉ, M.D.
Department of Obstetrics,
Gynecology and Human Reproduction
Institut Universitari Dexeus
C/Gran Vía Carlos III
Barcelona 08028 (Spain)
e-mail: clacol@dexeus.com