Ectopic pregnancies in a Caesarean scar: review of the medical approach to an iatrogenic complication

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Implantation of a pregnancy within a Caesarean fibrous tissue scar is considered to be the rarest form of ectopic pregnancy and a life-threatening condition. We conducted a computer search of the English literature of all studies since 2002 to gather updated data on the outcome of such pregnancies. Sixty-six new cases were reported since 2002, possibly reflecting the increasing number of Caesareans currently being performed as well as the more widespread use of the transvaginal scan allowing their earlier detection. Analysis of these women’s obstetric history revealed that those at risk for pregnancy in a Caesarean scar appear to have a history of dilatation and curettage, placental pathology, ectopic pregnancy, and IVF. Twenty-one out of 39 for which this information was available (54%) had undergone multiple (> 2) Caesareans and 13 had previous dilatation and curettage, which might also be an associated factor. We review and discuss the features of contemporary work-ups, including a high index of awareness, a detailed history and a skilful ultrasound examination for an early and accurate diagnosis. Healthcare professionals should be familiar with the possibility of untoward sequelae and how a modern work-up can help in guiding conservative options, thus reducing morbidity and preserving fertility.

Key words: pregnancy in scar/sonography/treatment

Introduction

Implantation of a pregnancy within a Caesarean fibrous tissue scar is considered to be the rarest form of ectopic pregnancy and constitutes a life-threatening condition (Fylstra et al., 2002). Placenta accreta (and its severe form placenta percreta) and placenta previa in which implantation occurs in the lower uterus are similar major complications in which trophoblastic tissues invade the uterine wall (Chazotte and Cohen, 1990). A pregnancy located in a Caesarean scar is, however, considered to be even more aggressive than placenta previa or accreta because of its early invasion of the myometrium, i.e. in the first trimester (Seow et al., 2000) (Figure 1). This is because of the very high risk for uterine rupture with all of its related maternal complications (Herman et al., 1995; Fylstra, 2002; Weimin and Wenqing, 2002).

Jurkovic et al. (2003) and Seow et al. (2004) have estimated that the prevalence of Caesarean scar pregnancy in their local population of women attending the early pregnancy assessment unit is ~1:1800 and 1:2216 respectively. Its true incidence, however, has not been determined because so few cases have been reported in the literature: only 18 cases appeared in the literature between 1978 and 2001 (Fylstra, 2002; Seow et al., 2004). We conducted a computer search of the English literature on all relevant studies since 2002 via MEDLINE (National Library of Medicine, Bethesda, MD, USA) and PubMed (National Library of Medicine, USA), and cross-referenced them to determine the report of these pregnancies. Medical subject heading search words used were: ‘Caesarean scar’ and ‘Ectopic pregnancy’. Only reports from peer-reviewed journals in the English language were included. We tallied 66 cases (including four case series) that had been added since 2002 (Table I). This value may reflect both the increasing number of Caesarean procedures currently being performed (Shennon, 2003), as well as the more widespread use of transvaginal scanning that enables an earlier detection of such pregnancies (Jurkovic et al., 2003).

Shih (2004) has recently reported that in his institute Caesarean scar pregnancy seems to be even more common than cervical pregnancy.

The current review aims to update our knowledge of, and experience with, the problematic issue of ectopic pregnancies located in Caesarean scars. The conditions related to the creation of this iatrogenic event, the difficulties in early diagnosis and the therapeutic challenges are discussed.
Predisposing factors for pregnancy in a Caesarean scar

Many theories have been proposed to explain the occurrence of this phenomenon. We consider the most reasonable one to be that the blastocyst enters into the wall through a microscopic dehiscent tract which may have been created through a trauma that occurred in association with a Caesarean or any other uterine surgery (Cheng et al., 2003), or even following manual removal of the placenta (Fylstra, 2002). IVF could represent a rare mechanism (Seow et al., 2000, 2004), even in the absence of any previous uterine surgery (Hamilton et al., 1992).

A Caesarean is known to present one of the risk factors for ectopic pregnancies and placental pathologies (i.e. placenta previa and placenta percreta) in the subsequent pregnancies (Hemminki and Merilainen, 1996). This was the case in two of our patients (Maymon et al., 2004) who had undergone either a cervical pregnancy or a placenta previa. A similar connection has also been described in other case reports (Haimov-Kochman et al., 2002; Salomon et al., 2003; see Table I).

Shinagawa and Nagayama (1969) found that an induced abortion preceded a cervical pregnancy in many cases. It follows that a similar trauma in the past may create conditions that favour implantation in that area. The same rationale may also apply to the Caesarean scar which can be a risky zone for implantation (Cheng et al., 2003), mainly when the scar is internally widened due to postoperative complications (Herman et al., 1995).

Because of the small number of cases and since no case–control studies using similar surgical technique for comparison have been reported, caution is needed when interpreting the current data.

Our survey revealed that 13 cases involved a previous dilatation and curettage (Table I). As had been the experience of Chuang et al. (2003); however, we could not determine the exact extent of this risk due to missing information in the case reports we analysed.

Jurkovic et al. (2003) found that 72% of their patients had undergone multiple (≥2) Caesareans. According to his opinion, this is another risk factor for inscar implantation of the subsequent pregnancy because of increased scar surface area (Jurkovic et al., 2003). Chuang et al. (2003) argued that the number of previous Caesareans does not appear to be a factor for such ectopic implantation. Our current survey found that 21 of the 39 (57%) parturient women for which this information was available underwent repeated Caesareans, which agrees with Chuang et al. (2003).

The high rate of Caesareans due to breech presentation and the subsequent occurrence of pregnancy in the resultant scar is another intriguing association that we encountered in five of our eight (63%) reported women (Maymon et al., 2004). Since this concomitant appearance was also described in another five reported cases (Neiger et al., 1998; Vial et al., 2000; Ghezzi et al., 2002; Hartung and Meckies, 2003; Yang and Jeng, 2003) (Table I), such an association might not be merely coincidental. Many of these operations (especially for breech deliveries) are currently elective procedures performed in a non-developed lower uterine segment, so that the healing processes following the operations might facilitate implantation of the blastocyst within the scar. We propose that the increasing number of Caesareans for various indications (Shennon, 2003) together with changes of the surgical technique (Maymon et al., 2004) and its indications (Maymon et al., 2004) might have some impact on the increase in inscar implantation. These possibilities need to be further examined.

Diagnosis

Since the clinical diagnosis of an early pregnancy implanted in a previous Caesarean scar can be very difficult, it may occasionally be delayed until the uterus ruptures and the patient experiences lifethreatening bleeding (Seow et al., 2000, 2004; Weimin and Wening, 2002; Yang and Jeng, 2003; Maymon et al., 2004). Thus, a prompt and accurate diagnosis is crucial. Diagnosis should be based on the pregnant patient’s history and her clinical manifestations, such as abdominal pain and any amount of bleeding (ranging from spotting to a lifethreatening haemorrhage). The most important investigation, however, is based on sonographic and Doppler flow findings (Marchiole et al., 2004).

Generally, sonography can detect an enlargement of the Caesarean scar in the lower segment and a mixed mass or a clear gestational sac that is attached to it. A very thin myometrium in a state of pre-rupture can occasionally be visualized inbetween the bladder wall and the gestational scar (Weimin and Wening, 2002).
### Table I. Published reports of Caesarean scar pregnancy

<table>
<thead>
<tr>
<th>Study (n = of cases)</th>
<th>Presenting symptoms</th>
<th>Primary diagnostic tool</th>
<th>Correct diagnosis at primary visit</th>
<th>Weeks of gestation at diagnosis</th>
<th>Primary treatment</th>
<th>Complications</th>
<th>Uterus preserved</th>
<th>Subsequent pregnancy report</th>
<th>No. of previous Caesareans</th>
<th>Indications for the previous Caesarean</th>
<th>Risk factors</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ghezzi et al., 2002 (n = 1)</td>
<td>NK</td>
<td>Ultrasound</td>
<td>+</td>
<td>7</td>
<td>Intramuscular 1 mg/kg body weight MTX + intra-amniotic KCl injection + bilateral uterine embolization</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
<td>1</td>
<td>Breech presentation</td>
<td>1 D&amp;C</td>
</tr>
<tr>
<td>Fylstra et al., 2002 (n = 1)</td>
<td>Light vaginal bleeding and mild low abdominal pain</td>
<td>Transvaginal ultrasound</td>
<td>+</td>
<td>7</td>
<td>Laparoscopy and laparotomy/hysterotomy</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
<td>2</td>
<td>NK</td>
<td>NK</td>
</tr>
<tr>
<td>Lam and Lo, 2002 (n = 1)</td>
<td>Suspected ectopic pregnancy</td>
<td>Transvaginal ultrasound</td>
<td>+</td>
<td>7</td>
<td>Multiple-dose intramuscular systemic MTX (1 mg/kg MTX and 0.1 mg/kg folinic acid)</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
<td>1</td>
<td>Pregnancy-induced hypertension</td>
<td>3 D&amp;C</td>
</tr>
<tr>
<td>Haimov-Kochman et al., 2002 (n = 2)</td>
<td>Slight vaginal bleeding</td>
<td>Transvaginal ultrasound</td>
<td>+</td>
<td>13 (missed abortion)</td>
<td>Intramuscular 50 mg/m² MTX Intramuscular 50 mg/m² MTX</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
<td>1</td>
<td>Placenta previa</td>
<td>3 D&amp;C</td>
</tr>
<tr>
<td>Pelvic pain</td>
<td>Transvaginal ultrasound</td>
<td>+</td>
<td>6</td>
<td>Full course of alternating MTX (50 mg/m²) and leucovorin</td>
<td>Xerophthalmia and mild mucositis</td>
<td>Yes</td>
<td>No</td>
<td>1</td>
<td>Fetal macrosomia</td>
<td>NK</td>
<td></td>
</tr>
<tr>
<td>Hartung and Meckies, 2003 (n = 1)</td>
<td>None</td>
<td>Transvaginal ultrasound</td>
<td>+</td>
<td>7</td>
<td>Intra-amniotic KCl injection</td>
<td>None</td>
<td>Yes</td>
<td>Yes</td>
<td>1</td>
<td>Emergency Caesarean</td>
<td>2 D&amp;C</td>
</tr>
<tr>
<td>Salomon et al., 2003 (n = 1)</td>
<td>None</td>
<td>Transvaginal ultrasound</td>
<td>Heteropathic pregnancy</td>
<td>6</td>
<td>Intra-amniotic KCl injection</td>
<td>None</td>
<td>Yes</td>
<td>Yes</td>
<td>1</td>
<td>Emergency Caesarean</td>
<td>2 D&amp;C</td>
</tr>
<tr>
<td>Chuang et al., 2003 (n = 1)</td>
<td>Severe vaginal bleeding</td>
<td>Ultrasound</td>
<td>+</td>
<td>7</td>
<td>Intraterine vasopressin and intramuscular MTX 1 mg/kg for four doses</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
<td>1</td>
<td>NK</td>
<td>1 D&amp;C</td>
</tr>
<tr>
<td>Yang and Jeng, 2003 (n = 3)</td>
<td>Severe vaginal bleeding after curettage Abdominal pain and vaginal spotting Vaginal spotting</td>
<td>Ultrasound</td>
<td>–</td>
<td>NK</td>
<td>Laparotomy/hysterotomy and transarterial embolization</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
<td>1</td>
<td>NK</td>
<td>2 D&amp;C</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Ultrasound</td>
<td>–</td>
<td>NK</td>
<td>Laparotomy/hysterotomy and transarterial embolization</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
<td>1</td>
<td>Breech presentation</td>
<td></td>
</tr>
</tbody>
</table>
Table I. Continued

<table>
<thead>
<tr>
<th>Study</th>
<th>Presenting symptoms</th>
<th>Primary diagnostic tool</th>
<th>Weeks of gestation at diagnosis</th>
<th>Primary treatment</th>
<th>Complications</th>
<th>Uterus preserved</th>
<th>Subsequent pregnancy report</th>
<th>No. of previous Caesareans</th>
<th>Indications for the previous Caesarean</th>
<th>Risk factors</th>
</tr>
</thead>
<tbody>
<tr>
<td>Weimin and Wengqing, 2002 (n = 15)</td>
<td>Light, painless vaginal bleeding</td>
<td>Doppler and ultrasound</td>
<td>7/15</td>
<td>12/15 received crystalline trichosanthin injected into the cervix + oral mifepristone or intramuscular MTX</td>
<td>3 cases total hysterectomy</td>
<td>12/15</td>
<td>No</td>
<td>NK</td>
<td>NK</td>
<td>NK</td>
</tr>
<tr>
<td>Jurkovic et al., 2003 (n = 18)</td>
<td>NK</td>
<td>Transvaginal ultrasound</td>
<td>4–23</td>
<td>Expectant management (3 cases) Local MTX or KCl (7 cases) D&amp;C + Foley catheter (8 cases)</td>
<td>8 cases of haemorrhage and blood transfusion</td>
<td>17/18</td>
<td>5 cases</td>
<td>13/18 previous multiple Caesareans</td>
<td>NK</td>
<td>2 IVF pregnancies</td>
</tr>
<tr>
<td>Maymon et al., 2004 (n = 8)</td>
<td>Silent picture (4 cases) Slight vaginal bleeding (2 cases) Missed abortion (2 cases)</td>
<td>Transvaginal ultrasound</td>
<td>7/8</td>
<td>1 case conservative follow-up 1 case aspiration 3 cases intra-amniotic MTX + systemic MTX</td>
<td>1 Caesarean hysterectomy</td>
<td>7/8</td>
<td>2 cases</td>
<td>4/8 previous multiple Caesareans</td>
<td>Breech presentation (5 cases) CPD (1 case) Fetal distress (2 cases) D&amp;C (3 cases) Cervical pregnancy (1 case) Placenta previa (1 case) IVF pregnancy (1 case)</td>
<td>1 IVF</td>
</tr>
<tr>
<td>Seow et al., 2004 (n = 12)</td>
<td>NK</td>
<td>Doppler and transvaginal ultrasound</td>
<td>10/12</td>
<td>D &amp; C and wedge resection (1 case) Local + systemic MTX (2 cases) Systemic MTX (1 mg/kg) Local MTX D&amp;C (one case)</td>
<td>Profuse maternal bleeding during D&amp;C The same woman had uterine rupture at 38 weeks of the subsequent pregnancy—the patient and the fetus died</td>
<td>11/12</td>
<td>2 cases</td>
<td>NK</td>
<td>Macrosomia followed by repeat section</td>
<td>NK</td>
</tr>
<tr>
<td>Shih, 2004 (n = 1)</td>
<td>Low abdominal pain and vaginal bleeding</td>
<td>Transvaginal ultrasound + 3D sonography and power Doppler Ultrasound</td>
<td>7</td>
<td>Laparotomy/hysterotomy</td>
<td>None</td>
<td>Yes</td>
<td>No</td>
<td>2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Marchiole et al., 2004 (n = 1)</td>
<td>Vaginal bleeding</td>
<td>No</td>
<td>9</td>
<td>Systemic MTX (100 mg) + D&amp;C + uterine artery angiographic embolization</td>
<td>Massive haemorrhage</td>
<td>Yes</td>
<td>Yes</td>
<td>1</td>
<td>Acute fetal distress</td>
<td>1 D&amp;C</td>
</tr>
</tbody>
</table>

NK = not known; MTX = methotrexate; KCl = potassium chloride; CPD = cephalo-pelvic disproportion; D&C = dilatation and curettage.
The most common sonographic criteria for a pregnancy in scar diagnosis are: (i) an empty uterus, (ii) an empty cervical canal, (iii) the gestational sac being located in the anterior part of the isthmic portion of the uterus with a diminished myometrial layer between the bladder and the sac (Godin et al., 1997; Seow et al., 2000, 2004; Fylstra, 2002), and (iv) a discontinuity in the anterior wall of the uterus being demonstrated on a sagittal view of the uterus when the direction of the ultrasound beam runs through the amniotic sac (Vial et al., 2000; Figures 2 and 3). These criteria assist in distinguishing this type of pregnancy from other diagnostic options, such as cervicoisthmic implantation, cervical pregnancy and spontaneous abortion in progress (Godin et al., 1997; Fylstra, 2002). In addition, a prominent peritrophoblastic flow is demonstrated around the gestational mass (Seow et al., 2000; Figures 2 and 3). One of the main advantages of the Doppler flow studies is the differentiation between a viable pregnancy in scar versus a non-viable intrauterine pregnancy (Jurkovic et al., 2003). This will obviously influence the choice of treatment. In cases of a non-viable intrauterine gestation, the gestational sac appears avascular, indicating that it has detached from the implantation site. In contrast, in a viable Caesarean scar pregnancy, the gestational sac appears well-perfused on Doppler studies (Jurkovic et al., 2003). The flow velocity around the gestational mass located in the scar is of low impedance (pulsatility index < 1) and of high velocity (peak velocity

Figure 2. Caesarean scar pregnancy at 7 weeks gestation. (A and B) Transvaginal and colour Doppler ultrasonography. A midline sagittal image demonstrating a gestational sac implanted at the isthmical region between the cervix and the empty uterine cavity (arrowheads), the anatomical location of a previous Caesarean scar. Note the prominent peritrophoblastic flow demonstrated around the gestational mass. (C) A midline longitudinal transabdominal scan demonstrating an empty uterine cavity and the scar pregnancy. The tip of the sac is bulging towards the bladder with a thin myometrium in between.

Figure 3. Caesarean scar pregnancy at 6 weeks gestation. (A) A midline sagittal transvaginal image demonstrating a gestational sac located at the isthmical region between the uterus and the cervix. (B) The same case with colour Doppler flow. Note the prominent peritrophoblastic flow around the gestational sac.
> 20 cm/s (Jurkovic et al., 2003). Others (Weimin and Wenqing, 2002) have reported a resistance index (RI) of the blood flow as being < 0.5 and a peak value ratio of systolic-to-diastolic (S/D) blood flow as being < 3.

We prefer to use transvaginal scanning for obtaining fine details of the gestation and its relation to the scar. We then conduct a meticulous abdominal scan with full bladder (Ravhon et al., 1997). This provides a ‘panoramic view’ of the uterus and an accurate measurement of the distance between the gestational sac and the bladder. The combined approach should confirm the diagnosis and provide additional information on the thickness of the myometrium between the bladder and the gestational sac. Others (Shih, 2004) have recently used 3-dimensional (3D) ultrasound and 3D Power Doppler. According to their experience, using the combination of the multiplanar views and 3D-rendered images permits more accurate diagnosis in the same situation.

In addition, the peritrophoblastic flow surrounding the trophoblastic shell may be further illustrated by 3D Power Doppler ultrasound to ascertain the diagnosis (Shih, 2004). Unlike Jurkovic et al. (2003), we try to avoid the ‘sliding organ sign’, which is defined as the inability to displace the gestational sac from its position at the level of the internal os by using gentle pressure applied by the transabdominal probe and transfundal manual pressure. We believe that this might provoke unnecessarily vaginal bleeding and even rupture in rare situations of a prerupture myometrium.

Some authors (Godin et al., 1997; Valley et al., 1998; Shufaro and Nadjari, 2001) have used magnetic resonance imaging (MRI) concomitantly with vaginal sonography as an additional diagnostic modality. The MRI transverse section image can clearly show the gestational sac embedded in the anterior cervix, but only when it is located in the outer surface of the vaginal canal (Weimin and Wenqing, 2002). It is our policy as well that of others (Nawroth et al., 2001) not to recommend MRI because we found that sonography combined with Doppler flow imaging are very reliable tools for diagnosing such cases. Others (Valley et al., 1998) have used cystoscopy to rule out any bladder penetration by the pregnancy. Roberts et al. (1998) described a Caesarean scar pregnancy as having a ‘salmon red’ appearance under endoscopy, and other clinicians have used hysteroscopy and laparoscopy for simultaneously visualizing and/or treating these pregnancies (Lee et al., 1999; Seow et al., 2000).

The vital importance of a correct diagnosis is highlighted in cases that were originally managed as an intrauterine pregnancy for which dilatation and curettage was performed and led to massive bleeding: at that stage, lifesaving laparotomy was mandatory (Seow et al., 2000; Yang and Jeng, 2003; Maymon et al., 2004).

Treatment modalities

Vial et al. (2000) proposed two different types of Caesarean scar pregnancies. The first is due to the implantation of the gestational sac on the scar with progression towards either the cervical-isthmic space or towards the uterine cavity. Such a condition may progress into a viable birth, but one with an increased risk of life-threatening massive bleeding from the site of implantation (Herman et al., 1995). The second is a deep implantation in a Caesarean scar defect with progression towards rupture and bleeding during the first trimester of pregnancy. Some authors (Ghezzi et al., 2002) believe that the difference between these two types of Caesarean scar pregnancy is of paramount importance. In the first case, the demonstration of a continuous connection to the cavum uteri justifies expectant management since pregnancy may continue until a viable birth. We adopted this policy in our first case (Herman et al., 1995) following a discussion with the couple and after reaching a mutual decision to implement expectant management. This pregnancy was uneventful and continued until 35 weeks gestation when an emergency Caesarean hysterectomy was performed to deliver a live infant weighing 3600 g. The patient required 16 units of blood because of persistent bleeding. Pathological findings revealed that the placental attachment in the lower segment had been lacking both decidua basalis and myometrium tissue and merely consisted of some connective tissues (Herman et al., 1995).

In the second type, the risk of late first trimester life-threatening bleeding is increased if immediate treatment is not undertaken.

Based on our experience (Maymon et al., 2004) and others' (Roberts et al., 1998), the prognosis for an uneventful term pregnancy is very poor. Therefore, our current policy is to recommend the termination of such a pregnancy once the correct diagnosis is made.

Although no treatment policy should be based on anecdotal reports because of the infrequent occurrence of uterine scar gestation, much is to be learned from each report (Haimov-Kochman et al., 2002; Jurkovic et al., 2003). In a haemodynamically stable patient, two principle management options may be considered, medical or surgical, both aimed to eliminate the gestational sac and retain the patient's fertility (Table I). In cases where the diagnosis is made early and the patient is haemodynamically stable, assessment of the thickness of the anterior uterine wall is essential (Ghezzi et al., 2002) because a non-surgical procedure is the most appropriate option when the trophoblast reaches the vesico-uterine space on the bladder wall, thereby obviating an extended operation. In such cases, the myometrium between the bladder and the gestational sac becomes thinner or even disappears due to distortion of the sac (Valley et al., 1998; Ayoubi et al., 2001). At that stage, only a thin serosal layer is left and sonographically detected measuring 1.2 mm (Godin et al., 1997) or < 2 mm (Weimin and Wenqing, 2002). Medical treatment consists of methotrexate (MTX) administered either systemically (Ravhon et al., 1997; Shufaro and Nadjari, 2001; Lam and Lo, 2002; Maymon et al., 2004; Seow et al., 2004; Shih, 2004), locally (Lai et al., 1995; Seow et al., 2004) or combined (Nawroth et al., 2001; Maymon et al., 2004; Seow et al., 2004). Because the Caesarean scar pregnancy is surrounded by fibrous scar tissue rather than a normally vascularized myometrium, some authors (Ravhon et al., 1997) have proposed that the systemic absorption can be limited, which may delay the absorption of the gestational sac, whereupon local MTX would take on greater importance (Fylstra, 2002). In addition, fine needle aspiration of the remaining fluid in the gestational sac may also be needed (Ravhon et al., 1997).

Others have used combined potassium chloride (KCl) injections directly into the fetal thorax and MTX injection into the sac

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and the surrounding myometrium (Godin et al., 1997). Local KCl injection alone was also applied (Hartung and Meckies, 2003; Salomon et al., 2003) and found to be a safe alternative, mainly in those extremely rare situations in which combined intrauterine and intrascar pregnancies are detected (Salomon et al., 2003). Another approach was introduced by Roberts et al. (1998) who successfully treated Caesarean scar pregnancy by the injection of hypertonic glucose directly into the sac, followed by systemic MTX administration. One should be aware that rupture of the scar and heavy bleeding may occur following medical treatment, as described by Jurkovic et al. (2003), or even 15 days after MTX treatment (Lai et al., 1995). This has led some authors to propose that the medical approach should be combined with either bilateral uterine artery embolization or vaso- pressin intracervical injection combined with 18 French Foley catheter balloon tamponade (Chuang et al., 2003), thus avoiding such complications (Ghezzi et al., 2002).

In one of our cases (Maymon et al., 2004) as well as in Jurkovic et al.’s (2003) experience, when the diagnosis was missed abortion, expectant management was chosen, and an uneventful outcome was achieved.

A recently described minimally invasive approach is that of endoscopic surgery (Lee et al., 1999). This includes hysteroscopy for visualization of the uterine cavity combined with incision and aspiration of the ectopic mass by operative laparoscopy. Aspiration of a very small gestational sac may facilitate pregnancy absorption (Ravhon et al., 1997; Jurkovic et al., 2003). Transcervical complete aspiration of the gestational sac under ultrasound guidance was performed without any complementary medical treatment only in one of our cases (Maymon et al., 2004). Jurkovic et al. (2003) performed dilatation and curettage (D&C) in eight cases and intraoperative haemorrhage followed in three of them. Similar experience faced Seow et al. (2004) in two cases. Therefore, D&C should not be considered the first choice of therapy (Shih, 2004). This is because the majority of the villi are implanted in the myometrium and it seems very unlikely that the gestational sac could be expelled by curettage without perforating the uterine wall or damage the urinary bladder, an accident that may cause lifethreatening bleeding and require emergency laparotomy (Fylstra, 2002; Weimin and Wenqing, 2002; Seow et al., 2004; Shih, 2004).

The surgical approach is supported by others (Rempen, 1997; Vial et al., 2000; Seow et al., 2000; Fylstra, 2002; Fylstra et al., 2002; Shennon, 2003; Shih, 2004) even in the presence of a non-bleeding patient. This includes elective laparotomy and wedge excision of the gestational mass when fertility is to be conserved. Several of these authors believe that even if recurrence is unlikely, the resection of the old scar with a new uterine closure can minimize the risk of recurrence (Fylstra, 2002; Fylstra et al., 2002). Wedge resection may, however, result in post-operative adhesions and fertility may be affected (Vial et al., 2000). In order to preserve fertility and reduce morbidity, surgery has been combined with selective embolization of the uterine arteries (Yang and Jeng, 2003).

We agree with Lee et al. (1999) that no single modality is entirely reliable and that none can guarantee uterine integrity. Treatment policy should be tailored to each patient and take into consideration the viability of the pregnancy and the gestational age as well as future family planning.

Post-treatment evaluation

Ravhon et al. (1997) have proposed that since the placenta is implanted on mainly fibrous tissue, absorption of the gestational sac is extremely slow. They reported that it took them >9 weeks to obtain complete clearance of the serum β subunit of hCG and only 3 months later did the transvaginal ultrasound show complete disappearance of the gestational sac and normal uterine anatomy.

A similar experience was reported by Donnez et al. (1997). Seow et al. (2004) have reported a range of 21–188 days for serum βhCG to reach undetectable levels.

Our follow-up protocol is similar to Jurkovic et al. (2003) who have proposed a weekly outpatient clinical assessment and measurement of serum hCG levels until they are undetectable. They performed an ultrasound examination to evaluate the size of the retained products of conception. This scanning examination was conducted on a monthly basis until no further pregnancy tissue could be detected. According to their experience, the hCG resolution time was between 6 and 10 weeks for patients who were treated with local MTX. Seow et al. (2004) have reported a range of 21–188 days for serum βhCG to reach undetectable levels.

In our experience, maternal recovery was complete following the original treatment protocol in all our three cases (Table I). There was no need for additional interventions and no side-effects related to MTX treatment were recorded (Maymon et al., 2004). However, Haimov-Kochman et al. (2002) reported one case of xerophthalmia and mild mucositis following a full course of alternating MTX and leucovorin.

Management of high-risk patients

We advise women who have had a pregnancy in scar and who are planning future pregnancies to have an early vaginal scan to confirm an intrauterine location of the new gestation (Maymon et al., 2004). Nowadays, with the advent of transvaginal sonography or with the use of saline infusion, uterine wall integrity could be detected even in the non-pregnant state (Monteagudo et al., 2001; Armstrong et al., 2003; Regnard et al., 2004). Caesarean scar defect, defined by the presence of fluid within the incision site (Armstrong et al., 2003), or any filling defect (‘niche’), defined as a triangular anechoic structure at the presumed site of a previous Caesarean scar (Monteagudo et al., 2001), might alert for uterine scar complication in the subsequent pregnancy (Armstrong et al., 2003; Jurkovic et al., 2003; Regnard et al., 2004). In addition, such a scanning approach will train the sonographer’s eyes to look for the scar in the pregnant uterus and to verify the integrity of the uterine wall. This is especially pertinent in the presence of an anterior gestational sac closely localized to the Caesarean scar.

Seow et al. (2000) reported detecting a scar defect by transvaginal sonography in their patient 4 years before she had an IVF-induced pregnancy in scar. This scan might be important for the subgroup of women at risk for pregnancy in scar, such as those with IVF pregnancies, previous ectopic pregnancies (including pregnancy in scar), previous placental pathologies and previous curettage or breech deliveries by a Caesarean. Specific
to IVF pregnancies in patients with a history of a Caesarean, Seow et al. (2000) recommended that the embryo should be transferred ≥4 cm from the cervical os, thus avoiding either Caesarean scar pregnancy or cervical pregnancy.

In our series (Maymon et al., 2004), two pregnant women have since spontaneously conceived an intrauterine pregnancy, and Jurkovic et al. (2003) have reported five subsequent pregnancies (Table I). Seow et al. (2004) had a patient who conceived again following evacuation of the pregnancy in scar by D&C. According to the patient request, she was followed until 38 weeks gestation when uterine rupture occurred. This ended in death of both the mother and the fetus. Nevertheless, those authors (Seow et al., 2004) do not favour laparotomy and suturing the scar to reduce such complications. However, they do recommend avoiding a pregnancy following such an event, probably for 12–24 months. Once a subsequent pregnancy has occurred they recommend elective Caesarean section as soon as the fetal lungs become mature. In one of our cases, a term Caesarean was performed because of fetal distress and there was no evidence of a previous pregnancy in scar. Salomon et al. (2003) reported removal of the gestational mass located in a Caesarean scar during the repeated Caesarean of a heterotopic intrauterine pregnancy.

**Summary**

Only 18 cases of pregnancy in scar were published in the English literature between 1978 and 2001 (Fylstra, 2002). Between 2002 and mid-2004 alone, however, the number rose to 66 reported cases. In view of the apparently increasing rate of Caesarean deliveries, healthcare professionals should be alerted to the possibility of this sequela. The old adage ‘Never allow the sun to set on a patient with a suspected ectopic pregnancy without taking her to surgery’ which is attributed to Louis M. Hellman, MD (R.W. Kistner; personal communication) is no longer a caveat that applies to all cases. This change in mandate may be particularly relevant to patients with pregnancy in scar. For the vast majority of those who were reviewed in this article, the sun rose and set for a few hospital days and then several weeks of outpatient care, mainly because the diagnosis was made when the patient was only mildly symptomatic and before rupture occurred, thus obviating major surgery.

In conclusion, modern work-ups enable an early and accurate diagnosis of pregnancy in a Caesarean scar, thus raising the diagnosis of scar pregnancy from an art to a science directed toward preserving fertility and reducing morbidity—a luxury beyond the reach of Dr Hellman. Because of the rarity of these cases, it is impossible to draw any firm conclusion as to the predisposing factors. Moreover, no universal treatment guidelines have been established to date (Fylstra, 2002), and there is no consensus on the treatment of choice. Still, it is widely agreed that it would be more prudent to interrupt such pregnancies as soon as a precise diagnosis has been made (Fylstra, 2002; Weimin and Wenqing, 2002; Maymon et al., 2004).

The gynaecologist must be prepared to deal with a very subtle challenge of deciding which is the best management option for the patient, and be familiar with the advantages and disadvantages of each one. Early detection and conservative treatment alone do not determine the ultimate outcome, and more emphasis should be placed upon reducing the known aetiological factors, which may contribute to the rapidly growing number of pregnancies implanted in a scarred uterus. Further studies are needed to compare different treatment methods in terms of their safety, reproductive outcome, optimal medication and dosage as well as economic feasibility.

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**References**


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