

Rupture of a caesarean scar ectopic pregnancy: A case report

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ABSTRACT

Background: Caesarean scar pregnancy is an uncommon form of ectopic pregnancy characterized by implantation into the site of a caesarean scar. Common clinical features include vaginal bleeding and abdominal pain; however, a significant proportion of cases are asymptomatic. The primary diagnostic modality is transvaginal ultrasound. There is no current consensus on best-practice management.

Case presentation: A 36-year-old woman, G7P2, presented to an early-pregnancy service with vaginal spotting and an ultrasound scan demonstrating a live caesarean scar ectopic pregnancy at 8 + 5 weeks' gestation. On examination she was hemodynamically stable with a soft abdomen. She was advised to have dilation and curettage (D&C) under ultrasound guidance; however, she was concerned that she might require more extensive surgery, such as a hysterectomy and so requested non-surgical management. On day 1 she underwent ultrasound-guided embryocide with lignocaine followed by inpatient multi-dose systemic methotrexate. Her beta-human gonadotrophic hormone level decreased. Repeat ultrasound on day 18 demonstrated a persistent caesarean scar ectopic pregnancy with increased vascularity, and so uterine artery embolization (UAE) was performed with a view to D&C the following day. This plan was altered to expectant management with ongoing follow-up by a different clinician who had had previous success with UAE alone. On day 35 the patient presented with life-threatening vaginal bleeding that required an emergency total abdominal hysterectomy.

Conclusions: Caesarean scar pregnancies are uncommon. Multiple treatment strategies have been employed, with variable degrees of success. Further research into risk stratification and management are needed to guide clinician and patient decision making.

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1. Introduction

Caesarean scar pregnancy (CSP) is an uncommon form of ectopic pregnancy associated with significant morbidity and mortality. CSP occurs when there is pathological implantation of a developing conceptus into the site of a previous caesarean section. The incidence of CSP is estimated at between 0.05 and 0.4% of all pregnancies and is expected to increase in prevalence in parallel with rising caesarean section rates [1,2].

The pathophysiology of CSP is not fully understood. A possible mechanism is that trauma caused by a caesarean section creates microscopic tracts through which an implanting blastocyst abnormally invades the affected myometrium [3,4]. The primary diagnostic modality for CSP is transvaginal ultrasound (TVS) and transabdominal ultrasound (TAS).

Supplementary magnetic resonance imaging (MRI) may be used in cases of diagnostic uncertainty.

Due to the rarity and heterogeneity of CSP no guidelines exist on best-practice management. A range of therapies have been described either in isolation or as part of combined management strategies [3]. Medical approaches include administration of intra-gestational sac methotrexate or long-course systemic methotrexate administration [3]. Surgical approaches may involve dilation and curettage (D&C), hysteroscopic resection, surgical excision via open, laparoscopic or transvaginal approaches, or hysterectomy [3]. Expectant management may be suitable for small, non-viable CSPs [3,5].

Here we present a case of a live CSP where surgical management was advised from initial presentation; however, this was refused due to patient preference. Management was initially with intra-gestational sac embryocide with lignocaine, followed by inpatient long-course systemic methotrexate. Uterine artery embolization (UAE) was then performed with a view to sequential D&C under ultrasound guidance. Following handover of patient care to a new clinician, who had had success with UAE alone, this plan was altered to ongoing expectant

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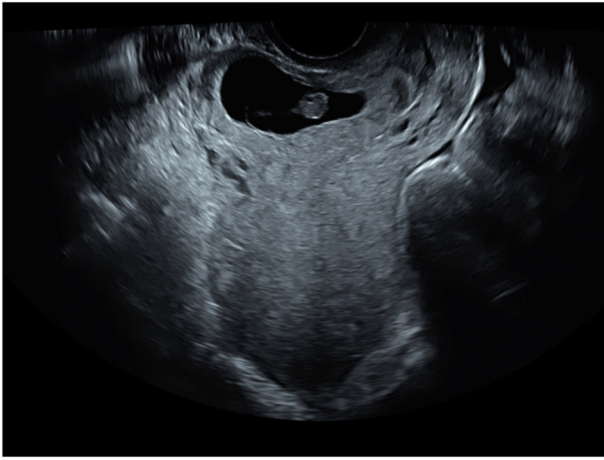


Fig. 1. Transvaginal image of caesarean scar ectopic pregnancy at initial presentation – mid sagittal view of the cervix and uterus.

management with outpatient follow-up. Ultimately, the patient required a hysterectomy for massive vaginal bleeding.

2. Case Presentation

A 36-year-old woman presented to an early-pregnancy assessment service (EPAS) with a history of vaginal spotting and an ultrasound scan demonstrating a live CSP at 8 + 5 weeks gestation. She had undergone two previous caesarean sections, three surgical terminations of pregnancy and two D&Cs for early miscarriage. Examination revealed a soft and non-tender abdomen. Serum beta-human chorionic gonadotropin (βHCG) was 88,445 IU/L. A repeat TVS confirmed the presence of a gestational sac containing a live embryo implanted at the location of the previous caesarean section, with a total pregnancy volume of 21 mL (Fig. 1).

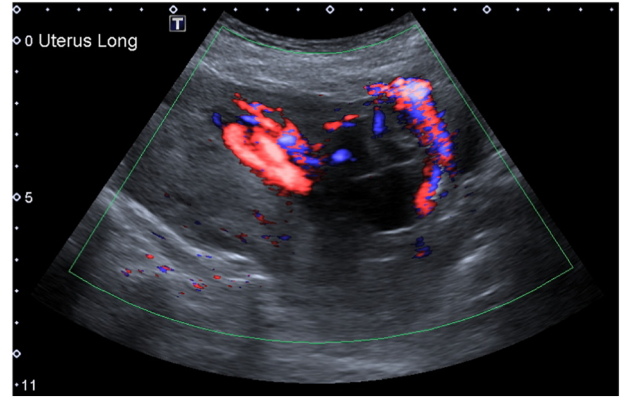
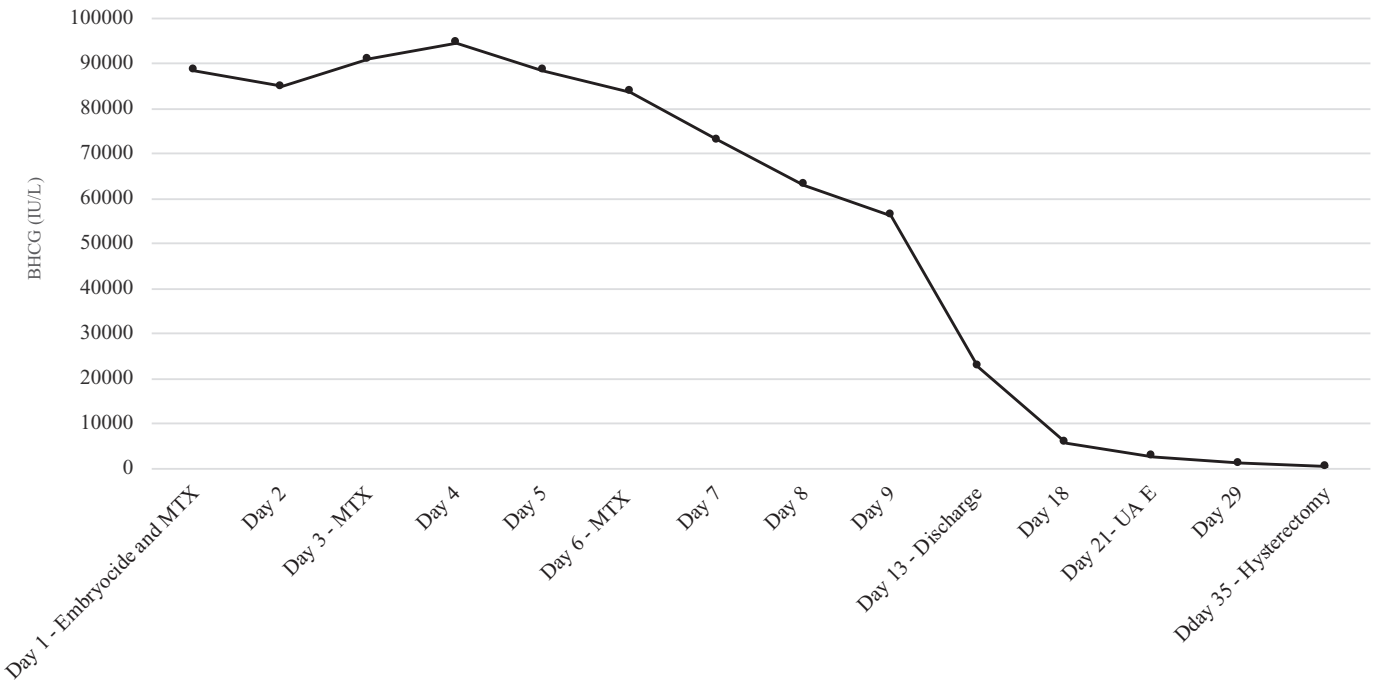


Fig. 3. Transvaginal ultrasound image with doppler demonstrating persisting caesarean scar ectopic pregnancy with vascularity.

The patient was informed that surgical options such as UAE followed by D&C or hysterectomy were recommended due to the significant risk of CSP rupture. Due to her strong preference to maximise fertility preservation she declined and opted for non-surgical management. She was counselled regarding the risk of massive vaginal bleeding necessitating emergency hysterectomy with this approach.

On day of presentation (Day 1) trans-vaginal ultrasound-guided embryocide with intra-cardiac lignocaine was performed. Inpatient systemic methotrexate using a three-dose, second-daily regimen (1 mg/kg IM) plus leucovorin rescue therapy was commenced the same day. After an initial rise in serum βHCG, levels steadily decreased to 22,785 IU/L by Day 13 (74% reduction from Day 1, trend shown in Fig. 2). A repeat TVS on Day 13 demonstrated a persisting CSP with a reduced total pregnancy volume of 8 mL. She was well with only light vaginal spotting and so was discharged with a plan for follow-up in EPAS with serial serum βHCG monitoring.

She was reviewed in EPAS on Day 18. The βHCG had fallen to 5822 IU/L (93% reduction from initial level) and she reported no vaginal



MTX = methotrexate. UAE = Uterine artery embolization.

Fig. 2. βHCG trend of caesarean scan ectopic pregnancy during management. MTX = methotrexate. UAE = Uterine artery embolization.

- 1) Empty uterus with clearly visualised endometrium.
- 2) Empty cervical canal.
- 3) Gestational sac within the anterior portion of the lower uterine segment at the presumed site of the caesarean scar.
- 4) Thinned or absent myometrium between the gestational sac and bladder.

Fig. 4. Proposed sonographic criteria for early diagnosis of caesarean scar ectopic pregnancies in the first trimester. Adapted from Osborn et al. [10].

bleeding. A TVS demonstrated a persistent CSP with an increased total pregnancy volume of 53 mL and associated peripheral vascularity (Fig. 3). The high risk of CSP rupture and significant bleeding were outlined once more and again surgical management was recommended. After some discussion she agreed to be readmitted for UAE followed by a D&C. The UAE was performed on Day 21 using Gelfoam. A conservative radiological approach was taken due to her fertility desires (some normal residual vascularity remaining on completion). As the initial clinician was on leave care was transferred to a new treating clinician. The planned D&C was cancelled in view of their previous clinical experience with UAE alone in successfully treating CSP. The woman was discharged from hospital with serial serum β HCG monitoring and EPAS follow up. She was seen on Day 29 in the EPAS and was asymptomatic with a decreasing serum β HCG of 1159 IU/L (99% reduction from Day 1).

On Day 35 the patient presented to the emergency department via ambulance in hypovolemic shock from massive vaginal hemorrhage. At the time of review she had ongoing vaginal bleeding with an estimated blood loss of 2.5 L. Her abdomen was soft and non-tender. Initial hemoglobin (Hb) was 88 g/L from a previous Hb of 115 g/L 6 days earlier. She was resuscitated before proceeding to an emergency laparotomy.

Intra-operatively the CSP was noted to have distorted the lower uterine segment, extending into the abdominal cavity but with intact overlying serosa. There was no intra-abdominal hemorrhage. Given the ongoing life-threatening vaginal bleeding the surgeon proceeded to perform an uncomplicated total abdominal hysterectomy. Total estimated blood loss was 3.25 L by the end of the procedure. Hb decreased to a nadir of 60 g/L post-operatively which required a total of 7 units packed red blood cells for replacement.

She had an uneventful post-operative course and was discharged home day six post-hysterectomy. Histopathology of the resected uterus confirmed embryonic tissue implanted within the lower uterine segment. She made a full recovery on reviewed 4 weeks later and was considering pursuing surrogacy to fulfil her fertility wishes.

3. Discussion

CSP is an uncommon and potentially life-threatening form of ectopic pregnancy. It is estimated to account for 0.15% of pregnancies after one caesarean and will likely increase in prevalence due to globally rising caesarean section rates [6,7]. Abnormal implantation of a CSP probably occurs through defects in the uterine tissue caused by the trauma of a previous caesarean section. The specific reasons they occur only in a minority of women is not clear [3].

The number of caesarean sections per woman does not appear to increase the risk of CSP, unlike the risks of abnormally invasive placentae, as an estimated 52% of cases of CSP occur after only one caesarean section [6]. IVF embryo transfer and previous caesarean section for breech presentation both appear to increase the risk of CSP, the latter possibly due to the need for a higher uterine incision in a poorly formed lower segment [6,8]. A review of 116 cases by Rotas et al the most frequent symptom of CSP was painless vaginal bleeding however 40% of patients were asymptomatic [6]. Average gestational age at diagnosis was 7.5 \pm 2.5 weeks [6].

Combined TVS and TAS with Doppler is the current diagnostic modality for CSP with a reported sensitivity of 86.4% [9,10]. Proposed

sonographic diagnostic criteria are outlined in Fig. 4 [10]. MRI can be used as an adjunct imaging modality to aid in decision making and operative planning through detailed characterization of CSP location, depth of myometrial invasion and presence of bladder involvement [11]. Subdivision of CSP based on location has been suggested. Type I (endogenic) grow toward the cervicoisthmic space or uterine cavity and may result in a viable pregnancy with high risk of placental site bleeding and morbidly adherent placentae [12]. Type II (exogenic) invade into the scar defect and progress toward the bladder and abdominal cavity and are associated with uterine rupture [12].

There is no consensus on best practice management of a CSP. A systematic review by identified 52 studies on management of CSP, four of which were RCTs and 48 of which were case series ($n = 2037$) [4]. Treatment plans included expectant management, systemic methotrexate, gestational-sac injection with methotrexate, D&C, hysteroscopic resection, transvaginal resection or laparoscopic resection, UAE, and repeated high-intensity focused ultrasound (HIFU) [4]. Expectant management ($n = 41$) has worst reported outcomes with 41.5% success rate and 53.7% severe complication rate [4]. UAE plus systemic methotrexate was performed in 427 patients with 68.5% success rate and 2.8% severe complication rate [4]. Surgical resection via a transvaginal approach has been described with a 99.1% success rate and 0.9% severe complication rate (6 studies, $n = 118$) [4]. HIFU followed by hysteroscopic suction curettage was described in one case series ($n = 53$) with a 100% success rate and no complications [13]. Generally medical treatments have slower resolution, persisting risk of rupture, and higher chance of additional treatment compared to surgical therapies [4]. Fertility outcomes after each intervention are unclear [4]. UAE has been associated with reduced fertility compared to myomectomy for the management of uterine fibroids, however the evidence is low quality and conflicting [14,15]. A conservative UAE approach was utilized in our case in an attempt to balance efficacy of the intervention and maximal future fertility.

This case highlights the severe risks associated with a CSP. Treatment options in this case were limited due to the patient's desire for minimally invasive interventions and maximal fertility preservation. Early intervention with UAE and sequential D&C may have removed the CSP and preserved fertility, however rupture leading to hemorrhage and hysterectomy may still have occurred with this approach. Due to the relative paucity of evidence into CSP management, clinicians are often guided by personal experience. Further research into treatment for CSP is needed to guide clinicians and patients in their decision making. Collaboration through registry of CSP cases and development of a high-quality multicenter prospective trial are both constructive future pathways. Investigation into fertility following the different treatment strategies is also of key interest.

Contributors

Harrison L. Odgers contributed to the design of the study, acquisition of information, and drafting of the manuscript.

Rebecca A. M. Taylor contributed to the design of the study and acquisition of information.

Jananie Balendaren contributed to the design of the study and acquisition of information.

Christopher Benness contributed to the drafting of the manuscript.

Joanne Ludlow contributed to the design and supervision of the study.

All authors saw and approved the final version of the manuscript.

Conflict of Interest

All authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

Informed consent was obtained from the patient.

Provenance and Peer Review

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