Case Report

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Subtotal Laparoscopic Hysterectomy for Management of Caesarean Section Scar Ectopic: A Case Report and Review of the Literature

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Abstract

Caesarean scar pregnancies are a rare form of ectopic pregnancy that are rarely compatible with foetal viability. Risks include rupture of haemorrhage resulting in substantial blood loss before viability, and abnormally invasive placenta in the very few pregnancies proceeding beyond the first trimester. Many cases have been reported, however, no clinical trials regarding treatment have been published due to the rarity of the condition. We believe this is the first described case of a subtotal (supra-cervical) laparoscopic hysterectomy to manage a caesarean scar ectopic pregnancy. Our method is a practical approach for management of caesarean section scar ectopic for patients where a total hysterectomy may not be possible or desired, offering rapid recovery and negligible blood loss compared to previously published open approaches.

Keywords: Caesarean scar pregnancy; Ectopic pregnancy; Sub-total laparoscopic hysterectomy

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Introduction

Caesarean scar pregnancies are a rare form of ectopic pregnancy that can be managed expectantly, medically or surgically and are rarely compatible with foetal viability [1]. Risks include rupture or haemorrhage resulting in substantial blood loss before viability, and abnormally invasive placenta in the very few pregnancies proceeding beyond the first trimester. Many cases have been reported, however, no clinical trials regarding treatment have been published due to the rarity of the condition. We believe this is the first described case of a subtotal (supra-cervical) laparoscopic hysterectomy to manage a caesarean scar pregnancy.

Case Report

We report on what we believe may be the first described case of caesarean section scar ectopic being managed by laparoscopic subtotal hysterectomy. A 44-year-old year old para 2 presented for a surgical termination of pregnancy at 7 weeks gestation by menstrual dates. A pre procedure ultrasound questioned the site of implantation. A second, more detailed ultrasound showed a live pregnancy measuring 23 mm × 21 mm × 12 mm, implanted within the previous caesarean section scar. The patient remained asymptomatic and was admitted to hospital for discussion and initiation of management. Haemoglobin on admission was 144 grams/litre (g/L) and beta human chorionic gonadotrophin (β -HCG) was 49613 international units per litre (IU/L).

Obstetric history included placenta praevia requiring an elective caesarean section, and an emergency repeat caesarean section where a uterine scar dehiscence was noted. Medical management at our institution includes multi dose intravenous methotrexate (50 mg/kg^2) in conjunction with direct injection into the gestational sac. We monitor therapeutic response with serial beta human chorionic gonadotrophin (β-HCG) concentrations and ultrasounds [2]. Cases are managed as inpatients at a tertiary facility due to the absence of a clear safety threshold and the requirement for immediate theatre access in the event of acute haemorrhage. Potential risks of medical management were explained to the patient including the side effects of nausea and vomiting, rash, photosensitivity, nephrotoxicity and oral ulcers, as well as the risk and management of uterine rupture or significant

haemorrhage.

The patient was able to consider a hysterectomy as she had completed her family prior to the current, unplanned pregnancy. The least invasive surgical approach, this being December the 23rd with a "home by Christmas" agenda, was laparoscopic hysterectomy and bilateral salpingectomy. She firmly expressed her desire for subtotal hysterectomy due to concerns about sexual function in her new relationship.

Laparoscopy identified an enlarged uterus consistent with the known gestation and otherwise normal pelvis. Vasopressin (desmopressin, 20 iU in 20 ml normal saline) was injected into the uterine fundus percutaneously using a spinal anaesthetic needle. Our standard laparoscopic hysterectomy technique was utilized commencing with bilateral salpingectomy and dissection of anterior and posterior leaves of the broad ligament, with reflection of the urinary bladder clear of the lower uterine segment. A combined bipolar/harmonic device (Thunderbeat, Olympus) was used in this instance. Using a 45 mm McCartney tube, the uterine vessels were secured and divided.

At this critical point the scar ectopic was readily identified and the level for uterine transection chosen: this was around 1 cm inferior to the ectopic, allowing for a 3 cm-4 cm length of cervix to remain. Residual cervical length was measured after transection, with a uterine sound. The cervical stump was covered with Surgical absorbable haemostat (Ethicon, USA) and peritoneum tacked over the cervix with 2.0 V-Loc™ (Medtronic, Minneapolis, USA) sutures to minimize transvaginal seepage.

The lower segment of the uterus, containing the pregnancy, was excised from the remaining specimen and removed in an Endocatch™ (Medtronic, Minneapolis, USA). The remainder of the uterus was morcellated using a LiNA Xcise™ (LiNA Medical ApS, Denmark) power morcellator under vision. Estimated blood loss for the whole procedure was 5 ml. Total anaesthetic time was 135 minutes. The patient made an uneventful recovery and was discharged 36 hours post-operatively **(Figure 1).** The patient's Day 1 post-operative haemoglobin level was 130 g/L.

Discussion

A caesarean scar pregnancy (CSP) is a rare form of ectopic

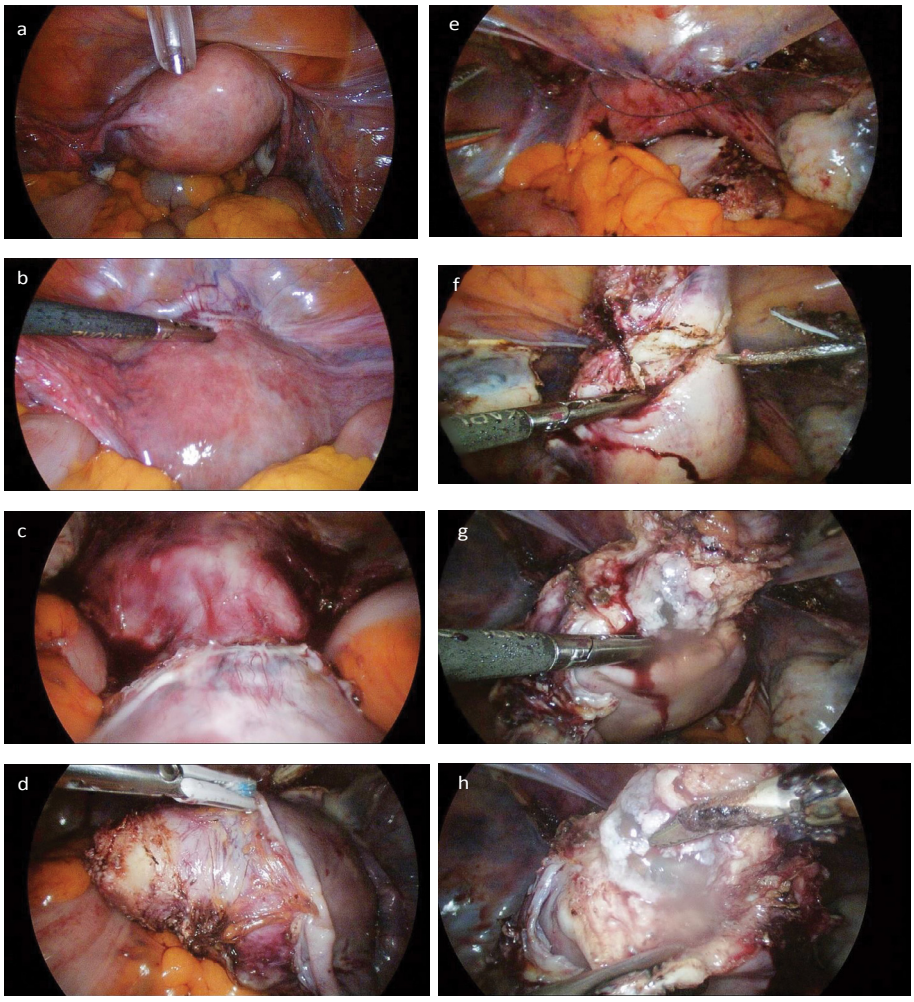

Figure 1 (a) Gravid Uterus (b) Caesarean section scar with ectopic (c) Completed SLH with *in situ* ectopic (d) Lower uterine segment with ectopic specimen (e) Peritoneum closed over the cervix (f-h) Dissection of scar pregnancy from main specimen.

pregnancy that occurs when the gestational sac implants in the myometrium of the uterus at the site of a previous caesarean section scar [3,4]. CSPs account for around 6.1% of ectopic, which represents 1 in 1800-2200 of all pregnancies [5]. Up to 72% of CSPs occur in women who have had 2 or more caesarean deliveries [6]. The incidence is thought to be increasing due to the global rise in caesarean delivery rate. CSP poses a significant risk of uterine rupture and severe haemorrhage and therefore hysterectomy and loss of fertility [7].

The main risk factor for a CSP is prior uterine surgery, particularly caesarean section or myomectomy. It is suggested that the risk of scar implantation may be proportional to the size of the uterine wall defect [8]. The exact pathogenesis of CSP is not fully understood, but may involve aberrant implantation [4].

CSP most commonly presents with painless vaginal bleeding or abdominal pain. Many remain asymptomatic and are detected at first trimester dating scans [9]. Early diagnosis enables prompt management and minimization of morbidity. Ultrasonography is the gold standard for diagnosis, with a reported sensitivity of 86.4% (95% CI 0.76-0.91) [10].

Sonographic criteria for diagnosis include: 1) An empty uterus with a clearly visualized endometrium, 2) an empty cervical canal, 3) A gestational sac within the anterior portion of the lower uterine segment at the presumed site of the caesarean scar; and 4) A thinned or absent myometrium between the gestational sac and bladder (<5 mm in two-thirds of cases) [6].

Management strategies are guided by case series, with a paucity of quality trial data.

Expectant management

Expectant management is associated with the highest maternal morbidity [7], particularly in the presence of foetal heart activity. Complications include uterine rupture, life threatening haemorrhage and eventual hysterectomy. Pregnancies without foetal heart motion are better candidates for expectant management [7]. Expectant management should only be offered to those patients who have minimal symptoms, a declining β-HCG, and who understand the potential risks of a ruptured ectopic pregnancy [9]. CSPs with foetal heart motion are associated with higher rates of first and second trimester complications as described above. Third trimester pregnancies may be complicated by an abnormally invasive placenta in 75% of cases [7]. In a series of 60 patients diagnosed between 5 and 14 weeks gestation, 48 had foetal heart motion at the time of diagnosis and four live births were reported [1].

Medical management

Options include systemic, multiple dose methotrexate, ultrasound guided intra sac injection or a combination of both [11]. Systemic methotrexate treatment follows the same dosing regimen as other non-tubal ectopic pregnancies and can be considered if the patient is stable, asymptomatic, with or without foetal cardiac activity, less than 8 weeks' gestation, a gestational sac of less than 2.5 cm and greater than 2 mm between the pregnancy and the bladder [11]. A systematic review in 2016 reported success in three quarters of 330 cases treated with systemic methotrexate [2]. Methotrexate is a folate antagonist that can potentially result in systemic side effects. These include acute kidney injury, vomiting, hepatotoxicity, neurotoxicity, myelosuppression, oral mucositis, increased photosensitivity and alopecia [12]. Systemic methotrexate is given as an intramuscular injection and uses a multi-dose regimen of (1 mg/kg^2) of methotrexate on day 1,3,5,7 and may be used in conjunction with leucoverin to decrease its systemic toxic effects [2].

Described protocols for ultrasound-guided local injection involve mechanically aspirating the gestational sac prior to injecting 50 mg of methotrexate, followed by saline flush. Injection of 2 ml (30 mmol/ml) of potassium chloride can also be used if a foetal heart beat is present [13]. Local injection, with or without KCl, may be successful in up to 80% of cases [4]. Some authors have suggested that the combination of intra-sac and systemic injection improves outcomes compared to a multi-dose protocol [9].

Medical management is often the treatment of choice as it is potentially fertility sparing and less invasive, however patients must accept a prolonged hospital stay and follow-up including serial β-hCG measurements and ultrasound monitoring [13,14]. After multi-dose methotrexate, we recommend daily serum β-hCG levels during treatment and pelvic ultrasound when the methotrexate administration is complete **(Figure 2).** Subsequent follow up requires a repeat ultrasound every two weeks until the ectopic mass has resolved. Serum β-hCG levels should be monitored from 1-3 times per week until negative [2].

Surgical management

There are a range of surgical approaches for management of CSP, ranging from minimally invasive methods to laparotomy. Management is based on the nature and acuity of the presentation and desire for future fertility. Surgical options include, blind dilation and suction curettage, hysteroscopy prior to dilation and curettage, resection of the CSP vaginally, uterine artery embolization, resection of the CSP via laparoscope or hysterectomy by any route [7].

Uterine preserving methods

Hysteroscopic management of CSP with resection of products under direct vision was first reported by Wang et al. in 2005 [15]. It was claimed that hysteroscopy allowed visualization of the gestational sac and associated vasculature which could be proactively secured by electrocoagulation [15]. The approach may invoke shorter theatre time and quicker return to normal duties. A Foley catheter may be used to provide tamponade [16] if required.

Uterine artery embolization (UAE) is a well described technique used for the treatment of uterine leiomyomata [17]. UAE in this context is claimed to reduce the immediate risk of hysterectomy, however subsequent miscarriage, preterm labour and postpartum haemorrhage may be increased. Risks include infection, premature ovarian failure and soft tissue ischemia; therefore we think UAE should not be used as first line treatment

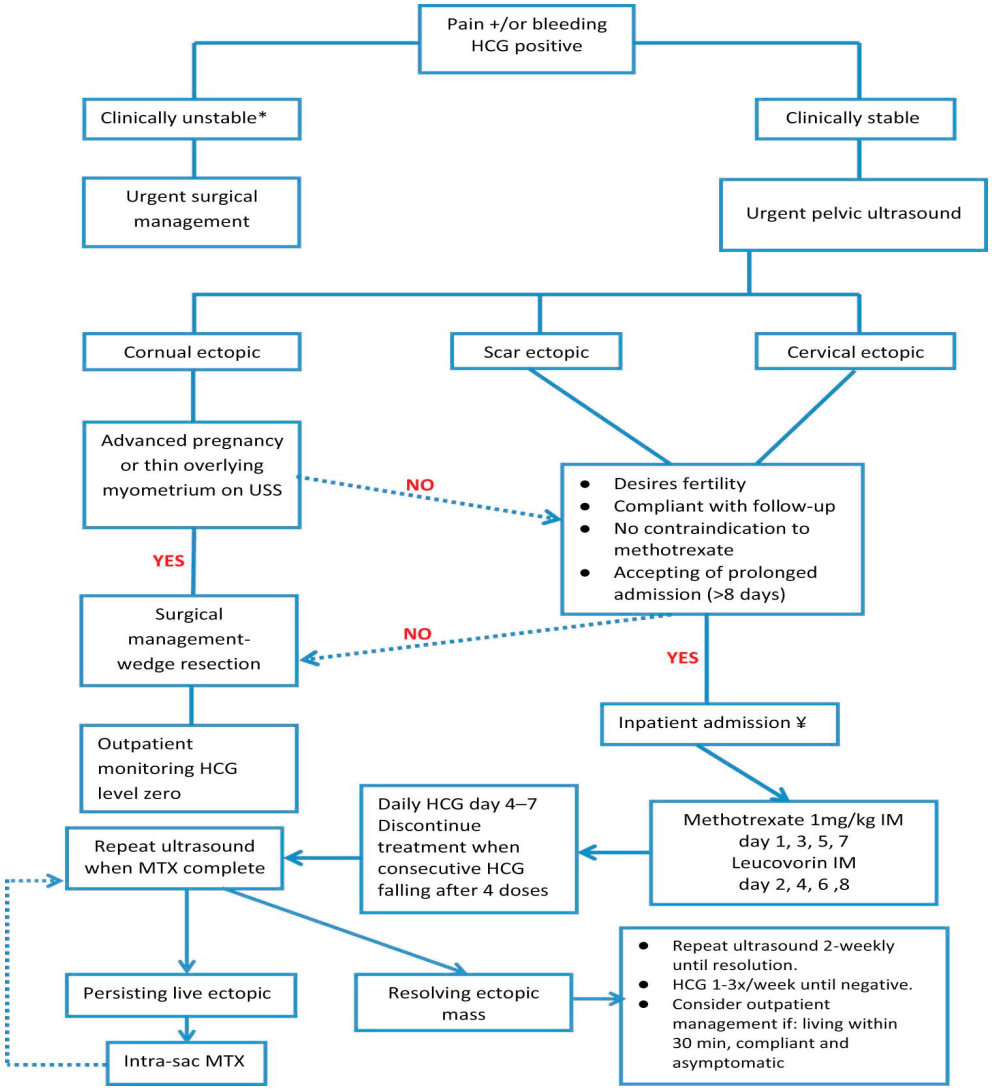

Figure 2 Suggested algorithm for non-tubal ectopic pregnancy management [2]. HCG: Human Chorionic Gonadotrophin; IM: Intra-Muscular; MTX: Methotrexate

***Clinical instability:** Systolic blood pressure <90 mmHg, haemorrhage requiring blood transfusion, postural blood pressure change >30 mmHg, syncope or presyncope or acute surgical abdomen (persisting severe abdominal pain or signs of abdominal peritonism). ¥ Patients from rural setting should be transferred to tertiary specialist centre.

[9]. UAE in combination with hysteroscopy with suction curettage has been described however the benefits are unclear [7].

Laparoscopy

Laparoscopic removal of a CSP may be considered for CSPs which are advancing anteriorly toward the abdominal cavity and bladder and less accessible by hysteroscopic approach [10]. Laparoscopy may involve wedge resection of the CSP and surrounding lower uterine segment. The bladder is dissected from the uterus, the CSP is excised and the uterine scar is repaired [11]. Additionally, vasopressin or bilateral uterine artery ligation may be used to minimize blood loss [17]. However, it has been suggested that occlusion of the uterine arteries may increase the risk of miscarriage in future pregnancies [18,19]. Reported complications of local resection include a conversion to laparotomy, massive haemorrhage and emergent hysterectomy.

Transvaginal resection

Le et al. described wedge resection via the anterior vaginal wall utilizing an approach not dissimilar to vaginal hysterectomy [14]. Compared to endoscopic surgery or uterine artery embolization, this small series appeared to result in a faster decline of serum β-hCG levels, no major complications and three subsequent pregnancies. Another series of 6 cases managed surgically with a transvaginal approach, reported an operating time between 45-80 minutes and serum β-hCG levels decreasing to normal limits within a month for all patients [15].

Laparotomy

Laparotomy is rarely the first choice for surgical management and minimally invasive approaches are often favoured. In women who have not responded to other treatments or present with

advanced gestation or complicated CSP, laparotomy with wedge resection and/or hysterectomy remain reasonable treatment options [10]. Compared to medical management, wedge resection or hysterectomy may reduce the risk of residual trophoblast and serum β-HCG levels usually return to normal limits within 1-2 weeks [10]. An open approach is suitable for suspected uterine rupture or when the myometrium between the ectopic pregnancy and bladder is greater than 2 mm [9]. Laparotomy usually leads to longer hospital stay, greater blood loss and the potential morbidity associated with open surgery [9].

Hysterectomy

One of the main goals of treating a CSP is fertility preservation. However, up to 12.5% of those with CSP eventually require hysterectomy, usually due to haemorrhage [5]. Hysterectomy may be performed vaginally, laparoscopically or via laparotomy. Decision for the route chosen mainly rests with the surgeon's skill set but is influenced by local resources and the patient's cardiovascular status [14]. Historically, claims have been made for the efficacy of the sub-total hysterectomy in preventing subsequent sexual and pelvic floor dysfunction. However, The Cochrane review [16] found no studies reporting a difference in sexual or pelvic floor function between total and subtotal hysterectomy [17-19].

Conclusion

Many surgical approaches for CSP have been described. Our centre's experience using a laparoscopic technique for subtotal hysterectomy appears to be a practical approach for patients where a total hysterectomy may not be possible or desired, offering rapid recovery and negligible blood loss compared to previously published open approaches. However, further studies are required to elucidate the safest management pathway for this rare form of ectopic pregnancy.

Disclosure Statement

The authors declare that they have no conflicts of interest and nothing to disclose.

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