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Diagnostic and management challenges of a rare case of caesarean scar pregnancy in a low-resource setting: a case report

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Abstract

Caesarean scar pregnancy is a rare type of ectopic pregnancy with the potential for catastrophic outcomes. A high index of suspicion is required for prompt diagnosis and intervention to improve outcomes. This report describes a rare case of Caesarean scar pregnancy, which was initially misdiagnosed as a threatened miscarriage and cervical ectopic pregnancy. A 35-year-old multiparous lady with two previous caesarean sections presented to the Gynaecology Unit of the Nnamdi Azikiwe University Teaching Hospital (NAUTH), Nigeria, at an estimated gestational age of 10 weeks, with recurrent vaginal bleeding of eight weeks' duration. She was referred to our facility from a private hospital, where she had first been managed as a case of threatened miscarriage and later as a cervical ectopic pregnancy. The transvaginal ultrasound in our facility was in keeping with a viable Caesarean scar pregnancy. The urine pregnancy test was positive, and the quantitative serum beta human chorionic gonadotropin was 75.6 mIU/ml. She had initial medical treatment with a combination of systemic multidose and intrauterine sac methotrexate and, subsequently, hysterotomy. Following systemic and local methotrexate, there was the demise of the foetus, which was evacuated at hysterotomy, and the uterine scar defect was repaired. She was discharged home in stable clinical condition one week after surgery. Her serum beta human chorionic gonadotropin dropped to 51.6 mIU/mL two weeks post-hysterotomy, and her urine pregnancy test became negative three weeks later. Though rare, caesarean scar pregnancy should be considered a differential diagnosis in reproductive-aged women with a previous caesarean section who present with vaginal bleeding in the first trimester.

Keywords

Caesarean scar pregnancy, ectopic pregnancy, medical treatment, transvaginal ultrasonography

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Introduction

An ectopic pregnancy (EP) is one in which the embryo implants outside the uterine endometrium.[1,2] It affects 1%–2% of pregnancies and is a leading cause of maternal morbidity and mortality in the first trimester, being responsible for approximately 9% of all pregnancy-related deaths.[3–5] While more than 95% of EP occurs in the fallopian tube, EP may uncommonly implant in the ovary (<3%), peritoneal cavity (0.9%–1.4%), cervix (<1%), or a previous caesarean section (CS) scar (<1%).[6]

Caesarean scar pregnancy (CSP) occurs when the embryo implants in the anterior lower uterine segment at the site of a

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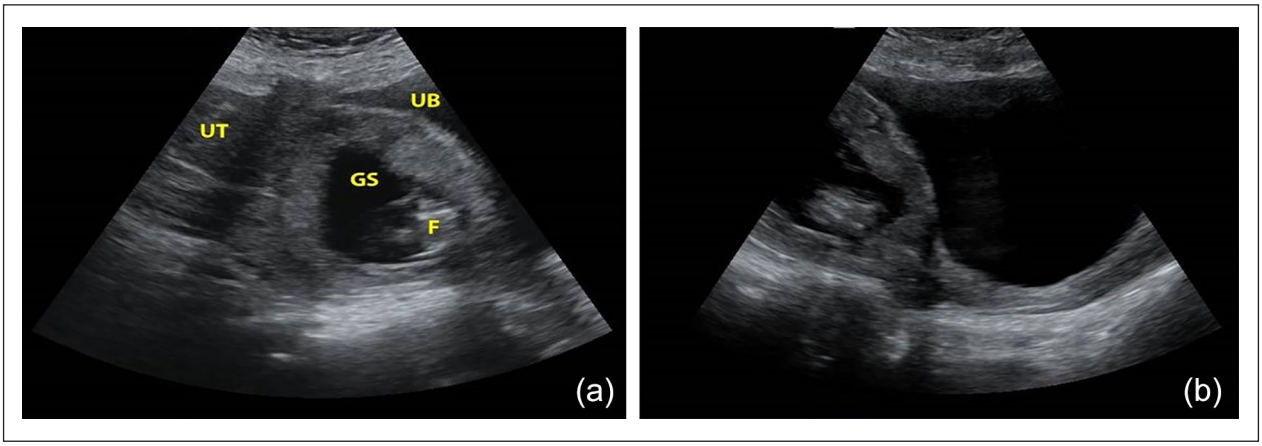

Figure 1. (a) A transabdominal ultrasound image shows the gestational sac in the lower uterine segment. There is no myometrium between the bladder wall and the gestational sac. The normal thickness of the myometrium is seen posteriorly. The placenta is attached to the caesarean section scar. A normal endometrial cavity is seen. (b) A transabdominal ultrasound image showing a normal cervical canal, excluding cervical ectopic.
F: Fetus; GS: Gestational sac; UB: Urinary bladder; UT: Uterus.

CS scar. It is the rarest type of EP, with an incidence of 1/1688–1/1800 of all pregnancies and 1:1800 to 1:2500 of caesarean deliveries,[7,8] representing 6% of all EP in women with a history of at least one CS.[6] Since the first case of CSP was reported by Larsen and Solomon in 1978, more than 1000 cases have been reported to date.[9,10] The increasing number of caesarean scar pregnancies is attributable to rising CS rates globally and advances in imaging.[11] We herein present a case of CSP in a 35-year-old multiparous Nigerian woman with two previous caesarean sections.

Case report

A 35-year-old gravida 3, para 2^{+0} presented to our facility at an estimated gestational age of 10 weeks with recurrent vaginal bleeding of eight weeks' duration. Bleeding was scanty, as she used one barely soaked perineal pad per day. There was no abdominal pain, dizziness, fainting spell or loss of consciousness, bleeding from other body orifices, or passage of vesicles per vagina. About four weeks prior to presentation at our facility (NAUTH), she had presented to another hospital, where she had a pelvic ultrasound scan (USS) done, which suggested she had a threatened miscarriage, and she was managed expectantly for a threatened miscarriage. A diagnosis of cervical EP was made on a repeat pelvic ultrasound at the same hospital two weeks later, for which she was referred to our facility for further/expert management.

The index pregnancy was spontaneously conceived. She had had two previous CSs. The first was an emergency CS 5 years ago, on account of failure of labour progress due to cephalopelvic disproportion (CPD), while the second was an elective CS two years ago, on account of one previous CS with bad obstetric history. She had no intra- or post-operative complications following both CS.

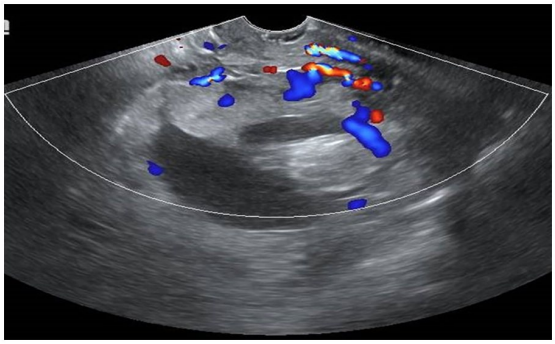

Figure 2. Colour Doppler ultrasound of the placental site on transvaginal US showing increased vascularity at the placental bed.

At presentation at our facility, general and systematic examination revealed normal findings. Abdominal and vaginal examinations were unremarkable, and the uterus was not palpable per abdomen. A transvaginal US (TVUS) and transabdominal USS done in our facility revealed an intact gestational sac at the level of the lower uterine segment, with the presence of foetal cardiac activity, the absence of myometrium between the bladder wall and the gestational sac, and an empty cervical canal, suggestive of a viable CSP (Figure 1(a) and (b)). Colour Doppler ultrasound of the placental site on TVUS showed increased vascularity at the placental bed (Figure 2). The urine pregnancy test (PT) was positive, and the quantitative serum beta human chorionic gonadotropin (β-hCG) was 75.6 mIU/ml. The full blood count and renal and liver function tests were all within normal limits. She was admitted and had medical management with methotrexate (MTX).

She received four doses of intramuscular (IM) 80 mg (1 mg/kg) MTX on days 1, 3, 5 and 7, with 8 mg (0.1 mg/kg)

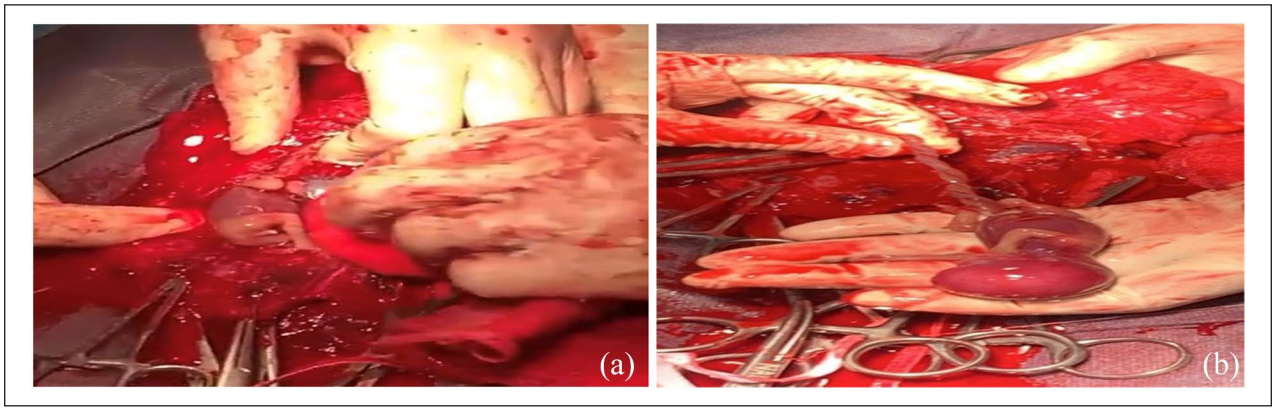

Figure 3. (a) Photograph taken at laparotomy showing the foetus with intact sac. (b) A non-viable foetus delivered in an intact gestational sac.

IM folinic acid rescue on days 2, 4, 6 and 8. Serum β-hCG after four doses of MTX was 83.6 mIU/ml. Repeat TVUS still showed a live CSP. She further had USS-guided intrauterine sac injection of 50 mg MTX. The gestational sac was localised under guidance using transabdominal ultrasound. The foetus was visualised to have gross body movement with good cardiac activity. The anterior lower abdominal skin was prepared with savlon and methylated spirit, and the proposed skin area was infiltrated with a local anaesthetic agent. Using a size 18-guage spinal needle, the sac was reached under ultrasound guidance. Amniotic fluid was aspirated to confirm the location, and 50 mg of MTX was administered into the sac. Intramuscular pentazocine was administered for pain control. A repeat TVUS 48 h after the intragestational sac MTX confirmed foetal demise with collapsed foetal skull bones. She subsequently had a hysterotomy. The hysterotomy was done via open laparotomy under spinal anaesthesia. Following standard skin preparation, the old pfanensteil scar was excised and developed into the peritoneal cavity following sharp and blunt dissections. Intraoperative findings were grossly marked adhesions involving the anterior abdominal wall and anterior uterine wall with dense fibrous bands; transverse anterior uterine wall dehiscence exposing foetal membranes; an intact amniotic sac containing a nonviable foetus; the right tube adherent to the body of the uterus, the left tube and both ovaries were grossly normal, the placenta was in close proximity to the bladder; however, there was no evidence of bladder infiltration; and estimated blood loss was 700 mls (Figure 3(a) and (b)). The uterus was closed in double layers, and the abdominal layers were subsequently closed in layers following standard surgical technique. The immediate postoperative condition was satisfactory.

Repeat quantitative serum β-hCG on the second and sixth postoperative days were, respectively, 96.01 mIU/ml and 58.8 mIU/ml. She was discharged home in stable clinical condition on the seventh postoperative day. Quantitative serum β-hCG repeated one week after discharge was 51.6 mIU/ml, and a urine PT repeated two weeks post-discharge was negative. A follow-up TVUS done following the surgery showed a normal uterus and a normal caesarean scar (Figure 4).

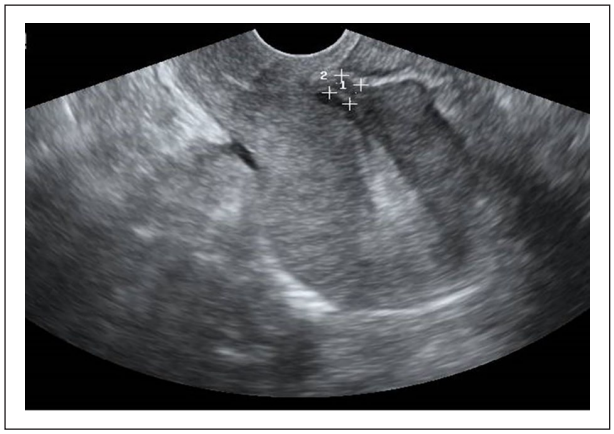

Figure 4. A follow-up post-surgery transvaginal US image showing a normal uterus and a normal caesarean scar, with calipers 1 and 2 demonstrating myometrial thickness and lower segment caesarean scar thickness.

Discussion

The pathogenesis of CSP remains uncertain. It is postulated that even though the majority of caesarean incisions heal without complications, due to the poor vascularity of the lower uterine segment, caesarean scars may heal improperly, resulting in microscopic dehiscent tracts (niches) that may predispose to trophoblastic invasion and implantation.[6,12] Factors that predispose to poor healing of a CS scar and consequently CSP include poor closure of the uterine incision, postoperative infection, and a short interval between a previous CS and subsequent pregnancy.[11,13] Others include preterm CS and CS done for non-progression of labour due to the need for a higher uterine incision, which is technically more difficult to close, with an associated risk of poor healing.[11,13]

Single-layer closure of the uterus with a non-inverting running suture has been reported to predispose to CSP due to impaired post-operative healing and defects within the scar.[14] Most CSP occur after one previous CS, and there is no clear correlation between the risk of CSP and the number of previous CS.[10,13] The index patient had two previous CS, with the first done for failure of labour progress due to CPD.

Transvaginal US is the main diagnostic modality, with a reported sensitivity of 84%.[13,15] The diagnosis of CSP by TVUS in the reported case was based on the USS diagnostic criteria first described by Godin et al., viz: (1) empty uterine cavity and cervical canal; (2) gestational sac within the anterior portion of the lower uterine segment at the presumed site of the caesarean scar; (3) thinned or absent myometrium between the gestational sac and bladder (<5 mm in two-thirds of cases); (4) peritrophoblastic low impedance, high velocity vascular flow on colour and a pulsed Doppler examination; (5) negative'sliding'organ sign'.[16] Based on imaging findings, there are two types of CSP. In type 1, or endogenic CSP, the implanted gestational sac grows towards the cervicoisthmic or uterine cavity, whereas in type 2, or exogenic CSP, which was the type this patient had, the gestational sac grows towards the bladder and abdomen, protruding anteriorly through the scar, with a greater risk of uterine rupture, bladder laceration, and massive haemoperitoneum.[6,10,15] Abnormal placentation can result in placenta praevia and placenta accreta/increta,[6] as was seen in our patient.

A high index of suspicion is required to diagnose CSP, as it is often misdiagnosed as cervical pregnancy or spontaneous miscarriage.[13] This was the case in the index patient, who was variously misdiagnosed as having a threatened miscarriage and cervical EP before referral to our facility. In cervical pregnancy, on USS, the uterus is empty and the gestational sac (which usually has a rounded configuration) is seen within the cervix with a closed internal cervical os, giving an hourglass appearance to the uterus with a ballooned cervical canal.[12] The gestational sac in a miscarriage is usually irregular and avascular, having detached from the implantation site, with an exponential fall in serum β-hCG level.[6,12] Vaginal bleeding is the most common presenting symptom in CSP.[13] In contrast to spontaneous or inevitable miscarriages, which usually begin with more extensive bleeding from the detached chorionic sac and cramping or lower abdominal pain, vaginal bleeding in CSP may be scanty, with only mild or moderate lower abdominal pain,[14] as in the index case. A third of patients may, however, be asymptomatic,[13] increasing the likelihood of a missed diagnosis.

There is currently no consensus on the optimal treatment and management of CSP.[12,13] The size and gestational age of the pregnancy, hemodynamic stability, and the patient's desire for future fertility will all influence the treatment plan that should be tailored for each patient.[17] There have been a number of therapeutic options used. Medical management, dilation and suction curettage, direct excision of CSP by an abdominal, laparoscopic, or hysteroscopic approach, and hysterectomy as the last resort have all been mentioned as options for management of CSP.[17]

Conservative medical management to preserve fertility is recommended in hemodynamically stable patients with unruptured CSP and options include systemic MTX, local embroycides or a combination of both.[14] Systemic MTX has a success rate of 71%–80%.[18] Given the short half-life of MTX, multi-dose regimens (which the index patient had) are more effective than a single-dose approach.[18] The likelihood of success is increased if the gestationl age (GA) is less than eight weeks old, with absent foetal cardiac activity, myometrial thickness of <2 mm, and a serum β-hCG level of <5000 mIU/ml.[12,18] Absorption of systemic MTX may, however, be limited by poor vascularization of the fibrous scar, necessitating additional treatment in a quarter of patients.[11] In such instances, local injection of embroycides, including MTX, potassium chloride, and hyperosmolar glucose, may be more effective.[11,18]

Surgical management is indicated in hemodynamically unstable patients, ruptured CSP, or when medical management fails, and options include hysteroscopy, laparoscopy, and laparotomy.[12] The index patient had a combination of systemic and local MTX and, subsequently, a laparotomy.

Other management options include preoperative uterine artery embolisation (UAE), followed by posterior isthmic hysterotomy and stand-alone UAE. There have also been reports of the application of Shirodkar cervical sutures, which produce a transient tamponade at the location of the ectopic implantation scar from a CS.[19] UAE has been shown to reduce the risk of subsequent haemorrhage in patients who undergo medical treatment or conservative surgery.[17] For women who need extra steps to ensure haemostasis, transcervical ultrasound-guided suction curettage combined with haemostatic stitching is an efficient treatment option for EP with a live caesarean scar.[20,21] It is linked to a minimal risk of hysterectomy and blood transfusion.[21] However, when gestational age grows and placental lacunae are present, the risk of significant intraoperative bleeding and the requirement for blood transfusion during or after surgical evacuation of live caesarean scar pregnancies increases.[20]

Conclusion

CSP, though rare, is a potentially catastrophic occurrence due to the increased risks of uterine rupture and massive haemorrhage. Its incidence is rising in parallel to the rising rates of CS globally and with advances in diagnostic imaging. A high index of suspicion is required to avoid a missed or delayed diagnosis. Prompt diagnosis and timely intervention are necessary to prevent life-threatening haemorrhage, preserve the uterus for future fertility, and improve outcomes.

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Author contributions

M.E.N., R.O.E., and A.C.I. performed the surgery, and manuscript writing. C.G.O, G.U.E., C.O.O., G.C.I., S.N.U., C.M.A., and A.F.N. were involved in manuscript writing. C.O.O. performed the ultrasound scan. All authors were involved in final proofreading, revision, and approval of the manuscript for submission to the journal.

Data and material availability statement

This is not applicable to this work, as no data was generated for the study.

Declaration of conflicting interests

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Ethical approval

This does not apply to case reports in our institution.

Informed consent

A written informed consent was obtained from the patient for the publication of this work.

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