International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Marwah S et al. Int J Reprod Contracept Obstet Gynecol. 2024 Aug;13(8):2119-2124
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20242081

Case Series

Pregnancy over caesarean scar site-account of four unusual cases in the form of a case series

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Received: 01 June 2024
Accepted: 02 July 2024

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ABSTRACT

Caesarean scar ectopic pregnancy (CSEP) is an ectopic pregnancy located in the lower uterine segment. Although it is one of the rarest types of ectopic pregnancy, the worldwide increasing rate of caesarean sections has made it important for young gynaecologists to be aware of the challenges posed by CSEP and its successful management. Doppler ultrasounds play a major role in its diagnosis. Early diagnosis is the key to a good outcome as untimely delay can lead to life-threatening haemorrhage, uterine rupture and irreversible loss of fertility. We present a case series of 4 patients of caesarean scar site ectopic pregnancy who presented to the gynaecology emergency and were treated successfully with various modalities depending on their patient profile.

Keywords: Ectopic pregnancy, Scar ectopic, Surgical excision

INTRODUCTION

Caesarean scar ectopic pregnancy (CSEP) is the rarest form of ectopic pregnancy reported worldwide (incidence=1:1800-1:2216). It's defined as pregnancy with a blastocyst implanting on a previous caesarean scar. However, a noteworthy escalation in incidence has been noted in the last three decades owing to the increasing rate of caesarean section, along with enhanced knowledge and grasp amongst obstetricians.[1] Delay in diagnosis can cause life-threatening bleeding/ uterine rupture, necessitating a hysterectomy leading to permanent infertility. Depending on the clinical presentation, management includes systemic methotrexate (MTX)/local uterine artery chemoembolization, dilatation and curettage, and excision of trophoblastic tissue (by laparoscopy/laparotomy) with uterine repair. There is a high likelihood that modern-day obstetricians encounter extra-uterine pregnancies in practice which become challenging due to limited accounts in literature, besides the absence of clear-cut management guidelines makes it imperative to equip budding gynaecologists with an adept understanding of early diagnosis and prompt management to avert associated morbidities.[2] Thus, a case series of successfully treated viable caesarean scar pregnancies is described, along with a pertinent literature review.

CASES SERIES

Case 1

A 32-year-old, G3P2L2 with a history of two caesarean sections (8 and 10 years back) presented to gynaecology emergency with two months of amenorrhoea and spotting for 2 days. No history of contraceptive use, abdominal pain or syncopal attacks.

The patient was haemodynamically stable with normal per abdominal examination. Per-speculum examination excluded local causes of vaginal bleeding. Bimanual pelvic examination revealed a bulky uterus with mild tenderness and fullness in anterior fornix and slight bleed.

Transabdominal sonography revealed an empty uterus and cervix with a viable gestational sac corresponding to 6 weeks gestation, lying in the anterior wall of the lower uterine segment, just above the internal OS.

The patient was monitored with β-hCG which was found to be 13374 mIU/ml. MRI pelvis showed a gestational sac-like cystic area attached to the thinned-out myometrium of the previous caesarean scar in the lower anterior uterine wall (with an absence of myometrium between the urinary bladder and gestational sac wall). These findings were suggestive of live CSEP.

Keeping in mind the possible complications and risk of bladder perforation the patient was taken up for laparotomy and wedge resection of CSEP. Per op dense adhesions were found between the urinary bladder and lower anterior uterine wall which were lysed, and bladder rent repaired. The gestational sac was excised from the scar site and the uterine tissue was repaired. Haemostasis was assured and bilateral tubal ligation was done.

The patient was discharged on 3rd post-op day with a self-retaining bladder catheter for 21 days. Histopathological examination confirmed the diagnosis of CSEP (Figure 1).

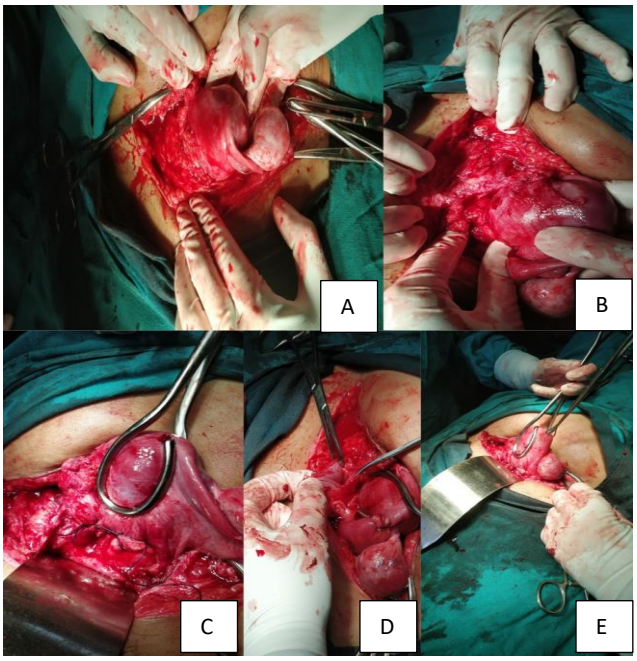

Figure 1 (A-E): Per operative findings of patient showing an adherent gestational sac arising from previous scar, with dense adhesions around in anterior abdominal wall; excised gestational sac followed by repair of scar, bladder injury at fundus and its repair.

Case 2

A 28-year-old, G3P1L1A1, with a history of caesarean section (3 years back), presented to gynaecological emergency with two months amenorrhoea and bleeding per vaginum for 2 days. Her past history was insignificant.

She was haemodynamically stable with unremarkable per abdominal examination. Per-speculum examination excluded local causes of vaginal bleeding. A bimanual pelvic examination showed a bulky uterus with fullness in the anterior fornix with mild tenderness. External OS was open and bilateral fornixes were free with bleeding observed on gloved fingers.

Transabdominal sonography revealed an empty uterine cavity with an inhomogeneous mass on the anterior wall of the uterus in the lower uterine segment without cardiac activity (aborting foetus/scar site ectopic).

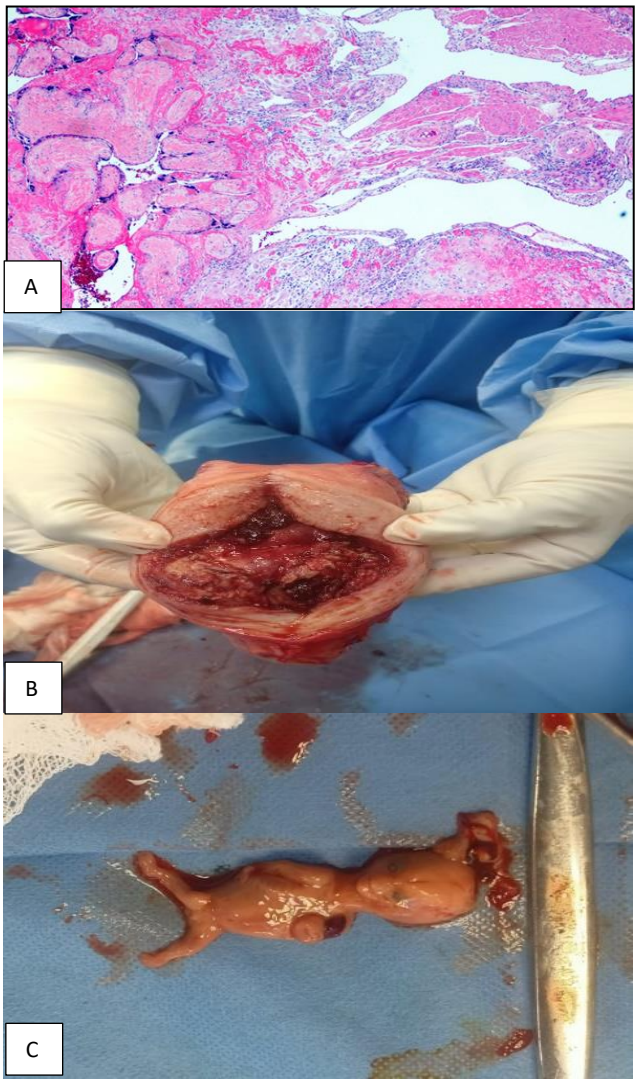

Figure 2 (A-C): H and E stain (100× magnification) shows immature chorionic villi adjacent to decidua. Smooth muscle bundles of uterine myometrium are located to right of decidua. Post hysterectomy uterus showing adherent placenta. Foetus of pregnancy implanted at the scar site

The patient was taken for emergency curettage which was converted to laparotomy because of ongoing bleed. Intraop, dense adhesions were present between bladder and uterus, and a vascular mass was seen abutting the previous caesarean scar. Attempts to separate mass from scar were unsuccessful due to continuous bleeding and a hysterectomy had to be done.

The patient was closely monitored and discharged on 5th postop day. Histopathological examination confirmed diagnosis of CSEP.

Case 3

A 25-year-old female, G2P1L0 with previous preterm LSCS presented to the gynae emergency at 10 weeks gestation (according to her last menstrual period), with painless vaginal bleeding since 1 day. Her menstrual cycles were regular with no significant past history.

The patient was haemodynamically stable with mild tenderness in the lower abdomen. Her per speculum examination was normal and per vaginal examination revealed a bulky uterus with free bilateral fornices.

Transvaginal sonography done on-site showed an empty uterine cavity with a gestational sac in the anterior myometrium invading the bladder serosa.

The patient was admitted and scanned by MRI on which a heterogeneously enhancing lesion of size 2.3×3×2 cm was seen with a fetal pole at the previous scar site in the anterior myometrium. The patient was keen to preserve her uterus for future pregnancy as she had no living child. Hence a medical management was planned along with an interventional radiologist for intrasac MTX administration. Serial monitoring of β-hCG was done and a decreasing trend was confirmed. However, the patient continued to have abdominal pain and spotting. Keeping future risk of scar ectopic pregnancy rupture in mind and patients' refusal for surgical intervention, uterine artery embolization (UAE) was performed after proper consent. The patient was monitored with weekly β-hCG and USG. The β-hCG levels normalised within 4 weeks. She was cautioned against a future pregnancy for the next 2 years along with possible complications.

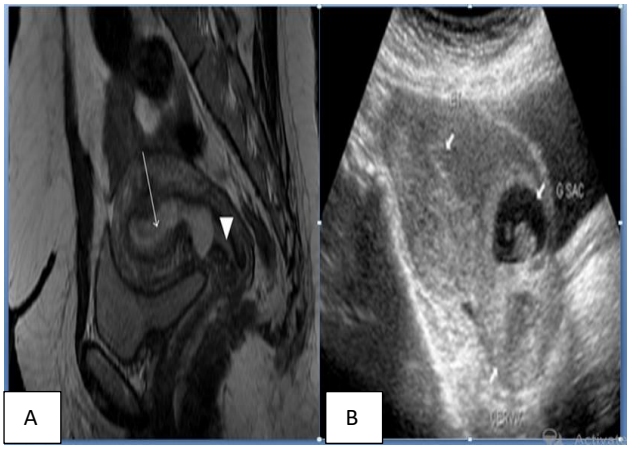

Figure 3 (A and B): Imaging of the patient; left picture depicting MRI T2 weighted image; right picture showing sagittal view of transabdominal depicting the gestational sac implanted at the lower pole of the uterus adjacent to the cervix (arrow), with Bladder being anterior to the scar on the uterus.

Case 4

A 30-year-old, G3P2L2 with previous one caesarean section presented to a gynaecology emergency at 6 weeks gestation with pain abdomen and spotting per vaginum.

The patient looked clinically pale (9 gm/dl) with a blood pressure of 110/70 and a pulse rate of 110/min. There were no significant findings on abdominal examination. Per speculum examination was normal and bimanual pelvic examination showed that the cervix was soft, and central with a 6-8 weeks sized uterus (bilateral fornices free with minimal bleeding per vaginum).

Transvaginal ultrasound showed a gestational sac in the lower uterine segment with a fetal pole and cardiac activity (scar site ectopic).

The patient was given the options of both medical and surgical management. The patient opted for intra-sac MTX administration followed by serial β-hCG level monitoring. The β-hCG levels showed a falling trend. The patient was counselled adequately regarding the risks of CSSE in future pregnancies and was advised to avoid conception for at least 2 years.

DISCUSSION

Ectopic pregnancy is the leading cause of morbidity and mortality in fertile women and is related to 4% of pregnancy-associated deaths.[3]

CSEPs are extremely rare, constituting 0.4% of all pregnancies and representing 6% of all ectopic pregnancies in patients with a case history of a previous caesarean section.[2]

Many theories have been postulated to explain the aetiology and mechanisms behind the occurrence of intramural extra-uterine gestation of which one implicates defective scar in the form of microtubular tract triggering scar implantation and another that the uterine scar is deficient in decidua basalis or contains a faulty layer of fibrinoid degeneration. Assisted reproduction techniques for pregnancy are the other ostensible risk factors.[4-9]

Two different types of CSEP have been illustrated-type I, implantation of gestational sac on uterine scar, progression towards cervical-isthmic space/uterine cavity is seen (away from the serosa), type II exhibits deep implantation into uterine scar, with progression towards serosa.[10]

The foremost presentation could be painless vaginal bleeding, mild lower abdominal pain or asymptomatic.[7,8,10-12] When silent, CSEP may cause perilous haemorrhage, uterine rupture, disseminated intravascular coagulation and even death.

Transabdominal/transvaginal sonography with Doppler has proven consistent in detecting such cases which evade

MRI.[6,10-14] Transvaginal sonography can be supplemented with transabdominal as the main modality for diagnosis based on the criteria described in GTG 21.[3]

The ultrasound diagnostic criteria for CSEP were defined as follows: Empty uterus with clearly visible endometrium, empty cervical canal, a gestational sac seen in the lower anterior uterine segment at the presumed site of the caesarean section incision scar, thin or absent myometrium between the gestational sac and the bladder (the majority of cases have a myometrium thickness <5 mm). "Sliding organ sign" and Doppler Flow present at the previous caesarean scar.

On pulsed Doppler examination, CSEP reveals flow waveforms of high velocity (peak velocity >20 cm/s) and low impedance (pulsatility index <1). Sonographically, the diagnosis is clinched by spotting a hysterotomy scar distended by an entrenched mass, with probable protrusion beyond the uterus anteriorly.[8,10-16] Additional observations include absent fetal parts in the uterine cavity, trophoblast sandwiched between the bladder and anterior uterine wall, deficient myometrium between the bladder and gestational sac, doppler confirmation of peri-trophoblastic vasculature perfusion, besides breach in the anterior uterine wall in sagittal plane.[8,10-16] One must attempt to delineate the extent of invasion into contiguous pelvic organs like the bladder through MRI when sonography is equivocal or inconclusive.

Expectant management places the mother at risk of an emergency hysterectomy if the pregnancy progresses beyond the first trimester.[8,10,14,17,18]

Medical modalities are preferred for asymptomatic women with <8 weeks gestation and myometrial thickness <2 mm between gestation sac and bladder.[19,20] These encompass systemic MTX administration, foeticide injection (like MTX, potassium chloride) directly into the sac, or a combination (foeticide followed by systemic drugs). Both single-dose and multidose protocols have been used. The standard single-dose regimen for MTX is 50 mg/m^2 IM whereas the multidose protocol consists of four doses of MTX 1 mg/kg given on days 1, 3, 5, and 7 with an alternating dose of folinic acid 0.1 mg/kg. CSEP with β-hCG levels <5,000 mIU/ml appear to respond best to systemic MTX.[3] However medical therapy has the shortcoming of slow resolution of pregnancy (up to months), with incessant possibility of rupture/ haemorrhage entailing hysterectomy. Also, repeated dosing of MTX, the requirement of a feto-maternal specialist for feticide, and long follow-up due to protracted (4-16 weeks) β-HCG normalisation (due to placental implantation on fibrous tissue), defy medical options.[19,20] Definitive options include wedge resection of ectopic pregnancy via laparotomy or laparoscopy, hysteroscopic excision, local injection of 5 mEq potassium chloride into the sac, and local/systemic MTX administration.[11,21-26]

Traditional uterine curettage (without additional therapy) risks critical haemorrhage, consequent to sac rupture and myometrial disruption.[18-26] High-intensity focused ultrasound (HIFU) combined with dilatation and curettage has also been exemplified as an effective modality.[27] UAE pre-treatment reduces the risk of subsequent haemorrhage and length of hospitalization in women undergoing conservative surgery or MTX injection.[8,19,20] A relatively novel approach is the use of hysteroscopy to confiscate CSEP under direct visualization, especially for type I, whereas laparoscopy can be utilized for a deeply implanted type II CSEP.[25,26] Transvaginal imaging can be supplemented with a laparoscopic approach. If on USG the anterior myometrial thickness is <3 mm, hysteroscopy can be preceded by laparoscopy to dissect the bladder peritoneum from the lower uterine segment in an attempt to remove the bladder from the surgical site of management and decrease the risk of injury. UAE circumvents the possibility of leaving residual trophoblasts reducing recurrence. Total abdominal hysterectomy has been purported to be an effectual solution in case of intractable haemorrhage/rupture.[4,6,8,14,17,18,23-28]

Table 1 shows the comparative evaluation of the present case series with previously published literature.

Table 1: Comparative evaluation of the present case series with previously published literature.

Authors, year	Presenting symptoms	Weeks of gestation	Key features	Management
Nankali et al, 2013[23]	Lower abdominal pain	11 weeks	Ruptured scar ectopic	Exploratory laparotomy followed by uterine evacuation and repair of uterine defect
Leite et al, 2014[7]	Asymptomatic	6 weeks 4 days	CSEP, with a live embryo,	Transvaginal ultrasound-guided injection of MTX, complemented with various doses of systemic MTX
Srinivas et al, 2014[11]	Painless spotting per vaginum	5 weeks	Live caesarean scar pregnancy	Mifepristone (200mg) followed by MTX
Sieczko, 2014[10]	Dark brown vaginal discharge and abdominal pain	6 weeks	Live pregnancy located in lower uterine segment within anterior wall, adjacent to a previous caesarean scar	Hysteroscopic resection of gestational sac done

Continued.

Authors, year	Presenting symptoms	Weeks of gestation	Key features	Management
Aich et al, 2015[12]	Asymptomatic	8 weeks	Live caesarean scar pregnancy	Laparotomy and hysterectomy
Schufreider et al, 2016[30]	Asymptomatic	7 weeks 3 days	Live caesarean scar pregnancy	combination of systemic MTX, intra-gestational sac potassium chloride (KCl) instillation, and transvaginal aspiration of gestational sac
Deepika et al, 2017[22]	Off and on bleeding per vaginum	6 weeks	Caesarean scar pregnancy with absent cardiac activity	Laparotomy with sac excision with scar repair
Roy et al, 2017[8]	backache, and mild vaginal bleeding	5 weeks	Live foetus in g sac over previous uterine scar	Injection of KCL into g sac along with local and systemic MTX injections
Ferdinando et al, 2020[31]	Stable, asymptomatic	5 weeks 6 days-11 weeks	Fetus with or without cardiac activity	Single dose MTX (50 mg intracavitary+50 mg IM) and folinic acid (15 mg/day for 30 days)
Fu et al, 2022[32]	Stable asymptomatic	≤10 weeks	Cardiac activity (+)	Expectant management was given at first and the ones with complications were then treated surgically as per protocol. A direct correlation was seen between myometrial thickness at scar site and complications.
Fatiha et al, 2023[33]	Bleeding per vaginum	6 weeks	Cesarean scar ectopic without cardiac activity	Laparoscopic excision followed by MTX based monochemotherapy
Mawan et al, 2023[34]	Severe lower abdominal pain and vaginal bleeding	12 weeks	Cesarean scar ectopic without cardiac activity	Laparotomy with excision and repair of rupture site
Present case series, 2023	Spotting per vaginum	weeks	Foetus with cardiac activity	Laparotomy with gestational sac excision with scar repair with bladder repair
	Bleeding per vaginum	≤10 weeks	Scar site ectopic without cardiac activity	Hysterectomy due to uncontrolled bleeding
	Bleeding per vaginum	10 weeks	Scar site ectopic with fetal pole without cardiac activity	Uterine artery embolisation
	Pain abdomen and spotting per vaginum	6 weeks	Foetus with cardiac activity	Intrasac MTX administration with βhCG monitoring

CONCLUSION

Management of CSEP is a challenging endeavour, due to its poor upshots. It thus requires proficient knowledge by obstetricians and radiologists, along with prudent assessment of potential differential diagnoses, and prompts individualized management, to forestall the associated maternal morbidity and mortality.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Marwah S, Agarwal A, Saini R, Balayan A. Pregnancy over caesarean scar site-account of four unusual cases in the form of a case series. Int J Reprod Contracept Obstet Gynecol 2024;13:2119-24.