

# Cesarean Scar Pregnancy: Diagnostic and Management Dilemmas in Low-resource Settings

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## ABSTRACT

**Background:** Cesarean scar pregnancy (CSP) is a rare but catastrophic event. The incidence of cesarean section (CS) is on an increasing trend and hence is the incidence of CSP. Failure to diagnose and initiate prompt management of CSP may lead to uterine rupture, massive hemorrhage, and even maternal death.

**Methods:** This study was conducted as a retrospective cohort study in women presenting to the Department of Obstetrics and Gynecology, MLN Medical College and Saket Maternity Nursing Home, Prayagraj, Uttar Pradesh, between January 2017 and February 2021, in which a final diagnosis of cesarean scar pregnancy was made. Comprehensive clinical, laboratory, and radiological data were collected from medical records of nine CSP cases. An analysis of demographic and clinical features with treatment modalities was done including age, gravidity, parity, number of previous CS, history of dilation and evacuation (D&E), history of abortifacient intake, presenting complaints, serum beta-hCG levels, sonographic features, and requirement for blood transfusion.

**Results:** The mean age of patients with a final diagnosis of CSP was  $30 \pm 4.5$  years. The previous history of D&E in this study was seen in 22% of patients. The majority (67%) of cases had one previous CS. The median duration from the last CS was 2 years. Approximately 55% of the patients had their last CS done without going into labor. History of abortifacient intake in present pregnancy was notably present in 67% of the patients. The mean gestational sac diameter in patients was  $15.4 \pm 4.0$  mm. Mean myometrial thickness between the bladder and gestational sac was  $2.2 \pm 1.18$  mm. Vascularity was mild in three and severe in five cases. The mean serum beta-hCG level at presentation was  $39891.6 \pm 36,305$  mIU/mL. The majority, i.e., five out of nine patients were managed surgically by laparotomy with a wedge excision of the scar and trophoblastic tissue followed by uterine repair.

**Conclusion:** High index of suspicion for the prompt and accurate diagnosis of CSP by both the gynecologist and sonologist is the need of the hour.

**Keywords:** Abortifacient, Abortions, Cesarean scar pregnancy, Cohort, Laparotomy.

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## INTRODUCTION

Cesarean scar pregnancy is a rare form of ectopic pregnancy in which the gestational sac is fully or partially implanted within the scar caused by a previous CS. It occurs in approximately 1 in 1,800–2,500 pregnancies.<sup>1</sup> With the dramatic increase in the number of CSs as well as advancement in ultrasound imaging quality, the frequency and diagnosis of CSP have increased significantly. Globally, the incidence of primary CS averages 18.6% of all births. In Asia, it has been reported up to 19.2%.<sup>2</sup> Failure to diagnose and initiate prompt management of CSP may lead to uterine rupture, massive hemorrhage, and even maternal death.

## MATERIALS AND METHODS

This study was conducted as a retrospective cohort study in women presenting to the Department of Obstetrics and Gynecology, MLN Medical College and Saket Maternity Nursing Home, Prayagraj, Uttar Pradesh, between January 2017 and February 2021. Including both the centers, a total of 16,642 deliveries were done during the study period with 5,824 deliveries by CS. Nine cases of CSP were diagnosed during the period. Diagnostic criteria on ultrasonography (USG) included an empty uterine cavity, empty cervical canal, and a clear gestational sac with or without cardiac activity or a mixed hematoma embedded in the anterior myometrium or fibrous tissue of the scar at the level of the isthmus with peri-trophoblastic vascularity. Myometrial thickness between the bladder and gestational sac was also noted. Comprehensive clinical, laboratory, and radiological

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data were collected from the medical records of nine CSP cases. An analysis of clinical and demographic features with treatment modalities was done including age, gravidity, parity, number of previous CS, history of dilation and evacuation (D&E), history of abortifacient intake, presenting complaints, serum beta-hCG levels, sonographic features, and requirement for blood transfusion.

## RESULTS

### Patient Characteristics (Table 1)

The mean age of patients with the final diagnosis of CSP was  $30 \pm 4.5$  years with gravidity and parity of  $2.5 \pm 0.49$  and  $1.3 \pm 0.47$ , respectively. The previous history of D&E in this study was seen in

**Table 1:** Patient characteristics

Case no.	Age (years)	Gravidae	Parity	Previous history of D&E	Number of previous LSCS	Last CS done in labor (Y/N)	Duration from last cesarean section/term/preterm	History of abortifacient intake (Yes/No)
1	36	3	2	No	2	Yes	8 years/term	Yes
2	26	3	1	Yes	1	No	18 months/preterm	No
3	26	2	1	No	1	No	7 months/term	Yes (1 month back)
4	25	2	1	No	1	Yes	2 years/term	Yes (at 6 weeks)
5	28	3	2	No	2	No	2 years/term	Yes
6	39	2 (IUI conception)	1	No	1	Yes	10 years/term	No
7	32	3	1	Yes	1	No	3 years/term	Yes
8	29	3	2	No	2	Yes	3 years/preterm	No
9	30	2	1	No	1	No	18 months/term	Yes

**Table 2:** Clinical characteristics

Case no.	Period of gestation (weeks)	Presenting complaints				Delay in diagnosis	Initial diagnosis on first ultrasonography	D&E done in present pregnancy
		Pain	Bleeding PV		Routine visit			
			Mild	Severe				
1	8	Yes		Yes	—	Yes	Incomplete abortion	Yes
2	9	No	Yes		—	Yes	Early intrauterine pregnancy	No
3	9	Yes		Yes	—	Yes	AV malformation	Yes
4	8	No	Yes		—	No	Scar pregnancy	No
5	9	No	Yes		—	Yes	Incomplete abortion	Yes (twice)
6	6	No	No		Yes	No	Scar pregnancy	No
7	7	No	Yes		—	Yes	Incomplete abortion	No
8	8	Yes	Yes		—	Yes	Early intrauterine pregnancy	Yes
9	8	Yes	No		—	No	Scar pregnancy	No

22% of patients. The majority (67%) of cases had previous one CS. The median duration from the last CS was 2 years. Approximately 55% of the patients had the last CS done without going into labor. History of abortifacient intake in present pregnancy was notably present in 67% of patients. The period of gestation at which the last cesarean was done was not clearly documented; however, seven patients were mentioned as term and two as preterm lower-segment cesarean section (LSCS) in the case sheets. Patients were not sure about the indication of CS and there were no documented operative notes available to analyze the indications of CS and techniques for the closure of the uterus.

**Clinical Characteristics (Table 2)**

The median gestational age at presentation was 8 ± 0.94 weeks. The most common presenting complaint was vaginal bleeding seen in seven (63%) out of nine cases followed by pain in 44% of patients. Only one-third of the patients were diagnosed as CSP in their first USG while two-thirds had a delayed or incorrect diagnosis before presenting. On earlier USG, three were diagnosed as incomplete abortion, two as early intrauterine pregnancy, and one as arteriovenous malformation. Forty-four percent of patients had D&E done in their present pregnancy for a spurious diagnosis before presenting. However, except for the first case in which there was torrential bleeding during D&E and a diagnosis of CSP could be

made only perioperatively, the rest of the patients were diagnosed correctly as CSP on repeat USG.

**Management Characteristics (Tables 3 and 4)**

Gestational sac diameter (GSD) was measurable only in five patients as one was taken up for emergency laparotomy directly without USG and three had hematoma formation at the implantation site, due to which it was difficult to measure the sac diameter separately (Fig. 1). The mean GSD in patients was 15.4 ± 4.0 mm. Embryonic cardiac activity was seen in four cases. Mean myometrial thickness between the bladder and gestational sac was 2.2 ± 1.18 mm. Vascularity was mild in three and severe in five cases. Mean serum beta-hCG level at presentation was 39891.6 ± 36,305 mIU/mL.

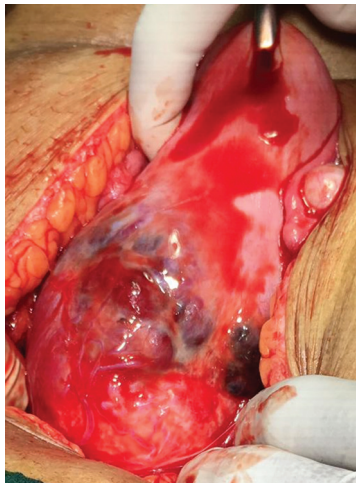
Treatment modalities used for nine patients in this study varied from medical therapy with systemic methotrexate to emergency hysterectomy. The majority, i.e., five out of nine patients were managed surgically by laparotomy with a wedge excision of the scar and the trophoblastic tissue followed by uterine repair in two layers with Vicryl number 1. Most of these patients presented after 8 weeks of gestation and had serum beta-hCG levels higher than 20,000 mIU/mL. The most common operative finding in them was a bulge of trophoblastic tissue at the scar site varying in size, degree of vascularity, and the presence or absence of any myometrial/serosal layer. The bladder was adherent to the scar covering the

**Table 3:** Management characteristics

Case no.	Hb%	Ultrasonographical features				Management			Blood/fresh frozen plasma transfused (units)
		Sac/crown-rump length (CRL)/hematoma size (mm)	Cardiac activity	Myometrial thickness between bladder and gestational sac	Vascularity	Serum $\beta$ -hCG level mIU/mL before treatment	Medical	Surgical	
1	7	-	-	No residual myometrium (perioperative finding)	-	-	-	Evacuation and curettage f/b Hysterectomy	3
2	8.5	Sac size 20 mm	+	No residual myometrium	Severe	30,100	Systemic methotrexate	Scar excision and uterine repair	2
3	6	Hematoma 56 x 55 mm	-	2 mm	Severe	16,670	Systemic methotrexate	Scar excision and uterine repair	5
4	11	Total sac size with hyperchoic trophoblastic tissue 57 x 36 mm, CRL 12.9 mm	+	3 mm	Severe	175,433	Systemic methotrexate	Scar excision and uterine repair	0
5	10.5	Hematoma 56 x 47 x 36 mm	-	4 mm	Mild	13,000	Multiple doses of systemic methotrexate, hematoma resolved completely in 6 months	No	0
6	11	Sac size 12 mm	Initially (+) but (-) after 1 week	5 mm	Mild	10,780	Mifepristone (oral) and misoprostol (vaginal) with systemic methotrexate	No	0
7	11	Sac size 10 mm	-	4 mm	Mild	8,150	Mifepristone (oral) and misoprostol (vaginal) with systemic methotrexate (lost to follow-up)	No	0
8	9	Sac size 15 mm	-	2 mm	Severe	21,000	Systemic methotrexate	Scar excision and uterine repair	2
9	11	Sac size 20 mm	+	No residual myometrium	Severe	44,000	Systemic methotrexate	Scar excision and uterine repair	0

**Table 4:** Composite table of clinical variables

Serial No.	Variables	Number (%)
1	Mean age (years)	30 ± 4.5
2	Mean parity	1.3 ± 0.47
3	Mean gestational age (weeks)	8.0 ± 0.94
4	Previous history of D&E	2 (22)
	Median interval from last CS (years)	2
5	No. of previous LSCS	
	1	6 (66.6)
	≥2	3 (33.3)
6	Type of LSCS	
	Emergency	4 (44.4)
	Elective	5 (55.5)
	Preterm	2 (22.2)
	Term	7 (77.7)
7	History of abortifacient intake	6 (66.6)
8	History of D&E in current pregnancy	4 (44.4)
9	Missed diagnosis	6 (66.6)
	Mean GSD visible in 5 patients (mm)	15.4 ± 4.0 mm
10	Myometrial thickness between the gestational sac and bladder (mean)	2.2 ± 1.18 (95% CI)
11	Serum beta-hCG level mIU/mL (mean)	39891.6 ± 36,305 (95% CI)
12	Surgical management	6 (66.6)
13	Medical management	3 (33.3)



**Fig. 1:** Cesarean scar pregnancy during laparotomy

ectopic gestation in two of the cases; however, none of them had any evidence of intraluminal invasion. The postoperative period was uneventful in all of them with adequate blood transfusion.

One patient went for an emergency laparotomy in view of the torrential hemorrhage and unstable hemodynamic status developed during D&E being done for incomplete abortion as suggested on USG. She was a 36-year-old lady with two live children and the last cesarean was 8 years back. Perioperatively,

a large hematoma (60 × 50 mm) with trophoblastic tissue was seen involving scar and adjacent myometrium almost invading the uterine serosa. Even though the bleeding was mild from this surface, a diagnosis of scar ectopic pregnancy rupture was made, and considering her hemodynamic status, continued vaginal bleeding, and completed family, the decision for hysterectomy was taken after informed consent.

Initial systemic methotrexate (50 mg/m<sup>2</sup>) was given to all the patients meanwhile preparing them for either surgical or additional medical treatment except the one who underwent a hysterectomy.

Three patients were managed medically after being counseled for prolonged follow-up and preparedness for necessary surgical intervention in case hemorrhage or inappropriate response was there. They were admitted initially for 24–48 hours and later as daycare during further follow-up. As shown in case 5, a 28-year-old lady with a history of D&E done twice in view of incomplete abortion presented at nine weeks of gestation. Albeit she had a hematoma of 56 × 36 mm at the scar site, considering her hemodynamic status, serum beta-hCG levels, and willingness to follow up, she was continued with medical management as weekly methotrexate (50 mg/m<sup>2</sup>) for four weeks. Hematoma size and vascularity took almost 6 months of time to resolve completely, and a more rapid fall in serum beta-hCG level was noted. Another patient for whom it was intrauterine insemination (IUI) conception after 10 years of the last CS did not give consent for any method of termination of pregnancy even after the early diagnosis of CSP at the first visit itself and agreed for termination only after the pregnancy became nonviable. After evaluation, she was given systemic methotrexate and mifepristone (200 mg) orally followed by a single dose of vaginal misoprostol (200 µg) after 48 hours, and fortunately, she passed the products of conception en masse after 12 hours. Although she did not get her serum beta-hCG levels done later, follow-up USG showed complete subsidence including vascularity in approximately 10 weeks. A third patient who was managed medically received only systemic methotrexate and mifepristone and became noncompliant and did not give consent for any further intervention with lost to follow-up (Table 3).

## DISCUSSION

The incidence of scar ectopic pregnancy is increasing globally with a rate of 0.15% in women with previous CS and 6.1% of all ectopic pregnancies.<sup>3,4</sup> The basic pathophysiology seems to be the invasion of blastocyst through a disrupted endometrium or a myometrial niche in a previous scar leading to the development of an embryo surrounded only by myometrium or fibrous tissue of the scar. The intramyometrial growth of the embryo explains the catastrophic behavior of this ectopic pregnancy.

Risk factors proposed for CSP include the number of previous CS, duration from the last CS, indication and timing of CS, technique of uterine closure, previous history of curettage, or manual removal of placenta. The mean age and parity of women in this study were slightly lower than the women studied in the systematic review done by Pektas in 2016; this may be due to early marriage and thus early obstetric carrier of women in this part of India. The previous history of D&E seen only in 22% of patients in this study is again not consistent with the study done by Michener et al., Australia, in 2009 in which all the thirteen patients had a history of at least one prior uterine curettage.<sup>5,6</sup> A definite correlation between the number of previous CS and the risk of CSP has not been established as most studies and also the present study have shown the occurrence of CSP even after only one CS.<sup>7</sup> Studies have shown that elective

cesarean delivery done for cephalopelvic disproportion, breech presentation, and fetal distress without women going into labor has an association with the occurrence of CSP.<sup>8</sup> Although we could not find out the exact indication in all the cases, almost 60% of the patients had last CS done at term without going into labor. It may be related to a higher uterine incision because of a poorly formed lower segment in such cases.<sup>9</sup> The median duration of 2 years from the last CS seen in this study is coherent with the study done by Michner, although other systematic reviews have shown an interval of less than 1 year.<sup>10</sup> No valid data are available in the literature regarding any possible association of history of abortifacient intake and CSP.

Median gestational age at presentation with vaginal bleeding and pain as the most common symptoms was comparable to most of the studies. As these clinical features mimic other common complications of early pregnancy like threatened and incomplete abortion, two-thirds of the cases in this study were initially misdiagnosed both clinically and sonographically which suggests that a routine early first-trimester USG should be cautiously done to make an early diagnosis in patients with a history of previous CS even if they are asymptomatic or seeking advice for a medical termination. Four patients underwent D&E in present pregnancy for a spurious diagnosis of either early intrauterine pregnancy or incomplete abortion; thus, the healthcare provider must mention the clinical history of previous CS in the requisition form and the sonologist must look for any features suggestive of CSP, differentiating it from low implantation, incomplete abortion, and intrauterine pregnancy. Considering the present era with the rising number of CSs and the potential of CSP to have higher complication rates with increased size and vascularity, sonographers and gynecologists should be familiar with the diagnostic criteria of CSP through different training programs.<sup>11</sup>

To date, there is no consensus regarding the optimal management of scar ectopic pregnancy. The aim is to remove the gestational sac before rupture as well as retain the future fertility with less chances of recurrence. Various modalities of management have been described in the literature depending upon factors like gestational age, clinical symptoms, hemodynamic stability, serum beta-hCG levels, myometrial thickness, availability of radiological and endoscopic expertise, fertility desires, feasibility of longer follow-up, as well as patient characteristics and wishes.

Medical management with systemic or local methotrexate alone or in combination has inconsistent success rates and is suitable only for hemodynamically stable patients with a gestational age of less than 8 weeks, absent cardiac activity, and hCG levels <12,000 mIU/L. Although they did not fulfill the proposed criteria for medical management, three patients in this study were managed medically as they were hemodynamically stable with hCG <20,000 mIU/L with mild trophoblastic vascularity and were willing for adequate follow-up.<sup>9,12</sup> Chances of failure and toxicity were informed. Successful use of combined methotrexate and mifepristone has also been shown in the study by Kalampos et al., in 2015; however, misoprostol has been used only for cervical priming in studies where hysteroscopic management was done, not for the sole purpose of CSP treatment (Fig. 2).<sup>13</sup>

USG-guided D&E combined with medical treatment has got variable success and complication rates in different studies and is again suitable only for hemodynamically stable patients with residual myometrium of at least >3.5 mm.<sup>5,14</sup> The risk of incomplete evacuation, heavy bleeding, uterine perforation, bladder injury, need for emergency laparotomy, salvage intervention, and

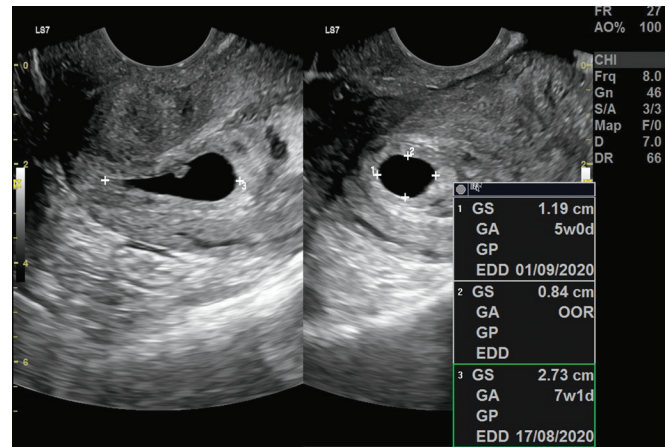


Fig. 2: USG image of CSP

recurrence associated with D&E in thinned-out myometrium was not acceptable for patients in this study. Moreover, the USG facility in the operating area was not available in both the setups. Although it requires an advanced setup with trained staff and equipment, laparoscopic or hysteroscopic resection of gestational tissue offers advantages of a minimally invasive procedure with quick recovery in a hemodynamically stable patient.<sup>9,12</sup> The expertise and operative equipment required for endoscopic management were not available in both the centers included in the study.

Surgical excision of scar and repair of the uterus by abdominal or laparoscopic approach offer the advantage of definitive management with quick recovery avoiding unpredictable, delayed, or failed response as well as inappropriate timing of complications associated with conservative methods. Also, it gives a better approach for the control of hemorrhage and repair of the myometrial defect. This was the most commonly employed modality in this study performed by abdominal route. An initial single dose of methotrexate was given to patients while being prepared for surgery to inhibit trophoblastic proliferation.<sup>5</sup>

A multidisciplinary approach involving a sonologist for USG-guided local injections or D&E may avoid surgery in appropriately selected patients and can be tried in low-resource settings too.

Limitations of this study were retrospective collection of data, small sample size, inappropriate documentation regarding previous surgery, and a very small number of patients receiving medical management.

## CONCLUSION

With the rising incidence of CS, keeping a high index of suspicion for the prompt and accurate diagnosis of CSP by both the gynecologist and sonologist is the need of the hour. Delay in diagnosis or misdiagnosis leads to high morbidity and mortality. Larger studies are needed to innovate the ideal modality; however, a multidisciplinary approach and substantial expertise for different individualized treatments should be there when considering conservative approaches. Patients should be emphasized about the long-term complications of recurrence, morbidly adherent to the placenta, and ruptured uterus, thus the need to report early in the next pregnancy. For primary prevention, the decision for CS without proper obstetrical indication on maternal request with inadequate trial of labor especially in primigravidae should always be reevaluated and the risk of future CSP should be informed in

such cases. More research to evaluate the relationship between different risk factors specifically related to the method of closure of the uterus in CSs is needed.

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