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Case Report

Management of cesarean scar ectopic pregnancies at an academic referral center: A case series☆,☆☆,★

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ABSTRACT

Objectives: To describe treatment and outcomes of patients with confirmed cesarean scar ectopic pregnancy (CSEP) at a tertiary referral center.

Study design: We reviewed a deidentified family planning clinical database for patients seen by our subspecialty service for CSEP from January 2017 through December 2021 in this case series. We extracted referral information, final diagnosis, management, and outcome measures including estimated blood loss, secondary procedures, and treatment complications.

Results: Of 57 cases referred for suspected CSEPs, 23 (40%) had confirmed diagnoses; one additional case was diagnosed during clinic evaluation for early pregnancy loss. Most (n = 50 [88%]) referrals occurred in the last 2 years of the 5-year study period. Of 24 confirmed CSEP cases, eight were pregnancy losses at the time of diagnosis. Fourteen cases were ≤50 days gestation or gestational size (7 [50%] pregnancy losses) and 10 >50 days gestation (range 39–66 days). We treated all 14 patients ≤50 days primarily with suction aspiration under ultrasound guidance in an operating room with no complications and estimated blood loss of 14 ± 10 mL. Of the 10 patients >50 days (maximum 66 days), seven were managed with primary aspiration of which five were uncomplicated. We treated one patient (57 days) had primary intrauterine double-catheter balloon with immediate hemorrhage requiring uterine artery embolization followed by an uncomplicated suction aspiration.

Conclusions: Patients with confirmed CSEPs at 50 days or less gestation or gestational size can likely be primarily treated with suction aspiration with low risk for significant adverse outcomes. Treatment success and complications are directly related to gestational age at treatment.

Implications: Ultrasound-guided suction aspiration monotherapy should be considered for primary CSEP treatment up to 50 days and, with continued experience, may be reasonable beyond 50 days gestation. Invasive treatments or those that require multiple days and visits, such as methotrexate or balloon catheters, are not necessary for early CSEPs.

1. Introduction

Cesarean scar ectopic pregnancies (CSEPs) represent less than 0.1% of all pregnancies and are associated with significant morbidity when the pregnancy is managed expectantly [1]. The diagnosis of CSEP is confirmed with transvaginal ultrasonography showing no gestational sac in the uterine body or endocervical canal, gestational sac in the lower uterine segment with anterior placental tissue located close to the prior hysterotomy, an absent or thin appearing myometrial layer between the gestational sac and the anterior uterine wall or bladder wall, and Doppler blood flow around the gestational sac [2]. CSEPs are described as type 1, or endogenic type (implantation occurs on the scar site and the gestational sac grows towards the cervicoisthmic space or uterine cavity), and type 2, or exogenic type (implantation in the scar and surrounding myometrium with progression to the bladder) typically based on growth patterns over time [3]. With type 2, a thin layer of myometrium, often < 5 mm, may initially be seen between the gestational sac and the bladder [4]. This thin myometrium commonly disappears as the pregnancy continues to grow, resulting in bulging of the gestational sac through the defect. Without treatment, CSEP results in placenta accreta spectrum with advancing gestation [4,5].

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Currently, no standard of care exists for optimal CSEP management. Expectant management of CSEP is not recommended due to high morbidity including life-threatening hemorrhage, uterine rupture, and hysterectomy [1,5,6]. A systematic review of CSEP treatment options describes medical management (methotrexate), surgical management (dilation and curettage [D&C], hysteroscopic, laparoscopic or vaginal CSEP excision, and hysterectomy), combined medical and surgical approaches, uterine artery embolization, and high intensity focused ultrasound ablation [7]. The authors reported surgical and combination treatments to be more effective than medication management alone; however, the number of cases in each of the 63 studies included in this review ranged widely, most with only 10 to 21 cases each [7]. Importantly, much of the older data does not specify D&C technique (sharp or suction). The authors made no specific recommendations about treatment options related to gestational age. More recently, CSEP treatment with double-balloon intrauterine catheter compression has been described in early gestations with good success, though data are limited by small case series or concurrent methotrexate use [8–10].

As a tertiary referral center, we evaluated our experience over a 5-year period to identify CSEP referral trends, treatment practices and outcomes by gestational age.

2. Materials and methods

We reviewed our University of California, Davis Family Planning deidentified clinical database for patients seen by our subspecialty service from January 2017 through December 2021 to find patients referred for or diagnosed with CSEP. The database includes referral and final diagnoses which allowed case identification. We extracted referral information, final diagnosis, ultrasound examination results, management, and outcomes for this case series. End points include estimated blood loss (EBL), treatment complications, and secondary procedures.

All care was provided or supervised by one or more of six Complex Family Planning subspecialists, usually in team consultation. All procedures for this diagnosis were typically performed by Complex Family Planning faculty or fellows. Standard practice during the time period was to perform an outpatient history, physical, and transvaginal ultrasonography to confirm diagnosis. Ultrasound findings for diagnosis included a gestational sac in the lower uterus and not in the endocervical canal with anterior implantation at the level of the lower edge of the bladder serosa and close to the prior hysterotomy site if visible. Subspecialists measured the myometrium, if present, between the outer edge of the hyperechoic implantation site and the uterine/bladder serosa.

Patients at 7 to 8 weeks gestation or less typically were scheduled for routine suction aspiration procedures in the operating room under transabdominal ultrasound guidance. Surgeons performed a paracervical block with a mixture of lidocaine hydrochloride 1% 20 mL and vasopressin 4 units. At 8 weeks or more, treatment commonly involved some other intervention prior to aspiration at the clinical discretion of the provider.

We used Fisher exact testing for statistical analysis. We received UC Davis Institutional Review Board exemption status. This case series is reported in accordance with the Preferred Reporting Of CasESeries in Surgery (PROCESS) Guideline [11].

3. Results

During this 5-year period, 57 patients had a referral or final diagnosis of CSEP, one of which had two referrals 4.5 months apart, for a total of 58 cases. Fifty-seven of these cases were referred for suspected CSEP of which 23 (40%) had a confirmed diagnosis (Table 1); the patient with two referrals had a CSEP both times. Most (n = 51 [88%]) referrals occurred in the last 2 years of the 5-year study period. The 34 referrals determined to not have a CSEP were diagnosed with a normal intrauterine pregnancy (n = 29), early pregnancy loss (n = 2), failed medication abortion (n = 2), and cervical ectopic pregnancy (n = 1). Overall, 45 referrals came from regional reproductive health clinics, most after presenting for abortion, of which 12 (26.7%) had a confirmed diagnosis. Conversely, non-reproductive health clinic sources had a confirmed diagnosis in 12 of 13 cases (92.3%, $p < 0.0001$).

Table 1
CSEP referrals to a University Family Planning Specialty Practice from 2017 to 2021

Year	# Referrals for CSEP	# Confirmed CSEP diagnosis
2017	2	2 (100%)
2018	1	0
2019	4	2 (50%)
2020	21	9 (43%)
2021	29	10 (34%)

CSEP, Cesarean Scar Ectopic Pregnancy.

The 24 total CSEP cases had a gestational age ranging from 39 to 66 days with 14 cases 50 days or less gestation or size (in the event of an early pregnancy loss) and 10 greater than 50 days on the day of treatment. Ultrasonography was completed by a university radiologist for seven cases, by a complex family planning specialist for 14 cases, and both specialists for four cases. Details of each CSEP are presented in Table 2.

All 14 cases at 50 days or less were treated primarily with suction aspiration under ultrasound guidance with a mean EBL of 14 ± 10 mL. Half (n = 7, 50.0%) had a diagnosis of early pregnancy loss. Eight had type 1 CSEPs and six had type 2 CSEPs. None had complications during or after the procedure.

Seven of the 10 cases at > 50 days gestation had primary aspiration, of which five were uncomplicated; one case required intrauterine balloon for bleeding (EBL 200 mL) postaspiration, and one was complicated by uterine perforation during primary aspiration with resultant exploratory laparotomy (EBL 250 mL). Two patients primarily received systemic methotrexate with plans for aspiration after significant human chorionic gonadotropin decline of which one was uncomplicated; the other also required an intrasac potassium chloride injection and, postaspiration, intrauterine foley balloon placement (EBL 250 mL). One patient (57 days) had attempted intrauterine double-catheter balloon under ultrasound guidance for primary treatment with immediate hemorrhage requiring uterine artery embolization followed by an uncomplicated suction aspiration (total EBL 650 mL with 500 mL following balloon insertion and 150 mL with aspiration) and transfusion of 1 unit of packed red blood cells.

4. Discussion

The goal of CSEP treatment is to resolve the CSEP expeditiously and safely while avoiding intraabdominal surgery, multistep or multiday procedures, uterine artery embolization, or intensive care unit admission. All 14 of our CSEP cases at 50 days or less gestation or gestational size were safely and successfully managed with suction aspiration monotherapy, avoiding the need for invasive or multistep procedures. Of the 10 cases with GA > 50 days, six were safely and effectively managed with suction aspiration (one after methotrexate pretreatment); the other four cases had complications including placement of intrauterine foley balloon or uterine artery embolization. Only two patients had clinically significant adverse outcomes, a patient with uterine perforation during aspiration at 52 days who had an exploratory laparotomy to ensure no resulting complications, and a patient at 57 days primarily treated with a double-catheter balloon who had immediate hemorrhage. These cases are a reminder of the potential for significant bleeding and morbidity that can occur with CSEP. A systematic review of

Table 2
Outcome measures from 24 confirmed cesarean scar ectopic pregnancies, 2017–2022

GA (days)[a]	EPL	G	P	# Prior CD	Myometrium (mm)[b]	Type[c]	Primary treatment	EBL (mL)	Complications
36	Yes	5	3	2	5	1	Suction aspiration	5	None
39	No	7	4	4	5.4	1	Suction aspiration	15	None
41	Yes	3	1	1	NM	1	Suction aspiration	30	None
41	Yes	4	3	3	1.9	1	Suction aspiration	10	None
42	Yes	7	3	2	3.8	1	Suction aspiration	10	None
43	Yes	4	2	2	2	2	Suction aspiration	5	None
43	Yes	5	2	2	0	2	Suction aspiration	5	None
44	Yes	4	3	2	4	1	Suction aspiration	10	None
46[d]	No	6	4	4	0	2	Suction aspiration	10	None
46	No	6	4	4	2.6	2	Suction aspiration	20	None
47	No	5	2	2	3	1	Suction aspiration	10	None
50[d]	No	7	4	4	4	2	Suction aspiration	25	None
50	No	7	3	3	3	1	Suction aspiration	40	None
50	No	5	2	1	1.1	2	Suction aspiration	5	None
52	No	4	3	3	1	1	Suction aspiration	200	Intrauterine foley balloon (inflated 20 mL) postaspiration for 6 h
52	No	8	4	4	2.5	1	Suction aspiration	5	None
53	No	2	1	1	4.2	2	Suction aspiration	250	Uterine perforation → exploratory laparotomy → intrauterine foley balloon (20 mL) for 22 h
53	No	4	3	3	4.5	1	Suction aspiration	20	None
54	Yes	8	2	2	3	1	Suction aspiration	10	None
54	No	7	3	2	3.4	2	Suction aspiration	10	None
54	No	4	3	3	2.3	2	Multidose MTX, suction aspiration 1 week after MTX	10	None
55	No	4	2	2	0	2	Suction aspiration	10	None
57	No	4	2	2	0	2	Intrauterine double-balloon catheter (10 mL/10 mL)	650	Hemorrhage with double-balloon catheter placement → UAE → suction aspiration, intrauterine foley (inflated 15 mL); transfusion of 1 unit packed red blood cells
61	No	4	3	3	1	2	Multidose MTX, KCl injection, suction aspiration (3 weeks after MTX),	250	Persistent cardiac activity after multidose MTX; intrauterine foley balloon postaspiration (inflated 55 mL) for 5 h

G, gravida; P, para; GA, gestational age; CD, cesarean deliveries; EBL, estimated blood loss; EPL, early pregnancy loss; MTX, methotrexate; KCl, Potassium chloride; NM, not measured; UAE, uterine artery embolization.
[a] Gestational age at initiation of treatment; gestational size if EPL.
[b] Ultrasonographic distance from edge of implantation site and bladder serosa.
[c] Cesarean section type [3].
[d] Same patient with two cases 4.5 months apart.

treatment options for CSEP found that D&C had a 76% success rate; however, the review did not stratify by gestational age [7]. As demonstrated in our results, success and complication rates were directly related to gestational age at treatment.

Our results are consistent with findings from a 2015 study of risk factors associated with successful D&C as primary treatment for CSEP, which showed a 91% (n = 173) success rate for gestations less than or equal to 48 days versus 55% (n = 49) success with CSEP greater or equal to 49 days gestation [12]. In that study, failure was defined as requiring further surgical or medical management (wedge resection, hysterectomy, or methotrexate with uterine artery embolization); successful cases included those with blood loss > 500 mL that were stopped by intrauterine Foley balloon compression [12]. We performed all cases in our series in the operating room setting due to concern of potential hemorrhage; however, it appears that for cases 50 days of gestation or less, office suction aspiration may be a reasonable option.

Timor-Tritsch and colleagues [8] described successful treatment of seven CSEP and three cervical ectopic cases at 43 to 53 days (median 48 days) gestation with double-balloon intrauterine balloon alone placed for 1 to 5 days (median 3 days). A separate study describes success using a single balloon foley balloon catheter used in 16 cases of CSEP (gestational age range 39 to 86 days, half of which were under 50 days) as part of multistep treatment with methotrexate and/or suction aspiration [9]. More recently, Kus et al. [10] reported successful treatment of 18 cesarean scar pregnancy and five cervical pregnancies with a double-balloon catheter, 87% of which also received systemic methotrexate. Although these techniques are novel and can be used for early CSEP, we have demonstrated that success is achievable with a simple aspiration procedure at these early gestations and does not require multiday and multistep treatments such as intrauterine balloon tamponade with or without methotrexate.

The increased number of referrals from the community with a < 50% confirmation rate of CSEP demonstrates the increased awareness of this diagnosis in the community and the recognition of the value of subspecialists for treating these cases. Most of the incorrect diagnoses came from regional reproductive health clinics which demonstrates heightened awareness and concern by practitioners in these facilities. Having appropriate referral centers when diagnoses are uncertain is important to ensure patient safety when a patient with a prior cesarean delivery presents for care. However, this finding also demonstrates an opportunity to improve ultrasonography skills for the practitioners at these centers.

Multiple existing case series describe CSEP treatment with lack of consistency in treatment approaches. The strength of our study is that we report our experience from a single referral center using a standard practice of primary suction aspiration for early CSEP cases. A weakness is a nonstandard approach for those cases over 50 days. Although this series represents a relatively large experience, a significant limitation is that the series is still relatively small.

Considering politics restricting safe reproductive health care, we urge our colleagues to continue using the nomenclature “cesarean scar *ectopic* pregnancy” rather than the frequently used “cesarean

scar pregnancy" to emphasize that these pregnancies pose life-threatening risks if managed expectantly [1]. Early screening for CSEP in any patient who has had a prior cesarean delivery is critical for safe management. The large increase in referrals for potential CSEP that we observed over the last 2 years of the study period likely reflects increased screening and awareness in our community.

Our data provide reassurance for primary treatment of patients with confirmed CSEPs at 50 days or less gestation or size (in the event of an early pregnancy loss) with suction aspiration with low risk for significant adverse outcomes. Those greater than 50 days may have more complicated outcomes but can likely still be treated with suction aspiration, although pretreatment with methotrexate and/or uterine artery embolism may be beneficial. The numbers in this case series are too small to be definitive about the gestational age at which primary aspiration should always be considered. We believe that, with continued experience, ultrasound-guided suction aspiration may be considered a rapid and safe treatment beyond 50 days gestation. For now, management of CSEP > 50 days gestation should be approached with the understanding of the potential for complications and unpredictable outcomes.

References

[1] Society for Maternal-Fetal Medicine (SMFM). Miller R, Gyamfi-Bannerman C. Society for Maternal-Fetal Medicine Consult Series #63: cesarean scar ectopic pregnancy. Am J Obstet Gynecol 2022;227(3):B9–20. https://doi.org/10.1016/j.ajog.2022.06.024

[2] Timor-Tritsch IE, Monteagudo A, Calì G, D'Antonio F, Kaelin Agten A. Cesarean scar pregnancy: diagnosis and pathogenesis. Obstet Gynecol Clin North Am 2019;46(4):797–811. https://doi.org/10.1016/j.ogc.2019.07.009

[3] Gonzalez N, Tulandi T. Cesarean scar pregnancy: a systematic review. J Minim Invasive Gynecol 2017;24(5):731–8. https://doi.org/10.1016/j.jmig.2017.02.020

[4] Jayaram PM, Okunoye GO, Konje J. Caesarean scar ectopic pregnancy: diagnostic challenges and management options. Obstet Gynaecol 2017;19:13–20. https://doi.org/10.1111/tog.12355

[5] Calì G, Timor-Tritsch IE, Palacios-Jaraquemada J, Monteaugudo A, Buca D, Forlani F, et al. Outcome of cesarean scar pregnancy managed expectantly: systematic review and meta-analysis. Ultrasound Obstet Gynecol 2018;51(2):169–75. https://doi.org/10.1002/uog.17568

[6] Zosmer N, Fuller J, Shaikh H, Johns J, Ross JA. Natural history of early first-trimester pregnancies implanted in cesarean scars. Ultrasound Obstet Gynecol 2015;46(3):367–75. https://doi.org/10.1002/uog.14775

[7] Maheux-Lacroix S, Li F, Bujold E, Nesbitt-Hawes E, Deans R, Abbott J. Cesarean scar pregnancies: a systematic review of treatment options. J Minim Invasive Gynecol 2017;24(6):915–25. https://doi.org/10.1016/j.jmig.2017.05.019

[8] Timor-Tritsch IE, Monteagudo A, Bennett TA, Foley C, Ramos J, Kaelin Agten A. A new minimally invasive treatment for cesarean scar pregnancy and cervical pregnancy. Am J Obstet Gynecol 2016;215(3):351.e1–8. https://doi.org/10.1016/j.ajog.2016.03.010

[9] Timor-Tritsch IE, Cali G, Monteagudo A, Khatib N, Berg RE, Forlani F, et al. Foley balloon catheter to prevent or manage bleeding during treatment for cervical and cesarean scar pregnancy. Ultrasound Obstet Gynecol 2015;46(1):118–23. https://doi.org/10.1002/uog.14708

[10] Kus LH, Veade AE, Eisenberg DL, Dicke JM, Kelly JC, Dickison SM. Maternal morbidity after double balloon catheter management of cesarean scar and cervical pregnancies. Obstet Gynecol 2022;140(6):933–5. https://doi.org/10.1097/AOG.0000000000004977

[11] Agha RA, Sohrabi C, Mathew G, Franchi T, Kerwan A, O'Neill N. PROCESS Group. The PROCESS 2020 guideline: updating consensus Preferred Reporting Of CasESeries in Surgery (PROCESS) guidelines. Int J Surg 2020;84:231–5. https://doi.org/10.1016/j.ijsu.2020.11.005

[12] Wu XQ, Zhang HW, Fang XL, Ding H, Piao L, Joseph Huang S. Factors associated with successful transabdominal sonography-guided dilation and curettage for early cesarean scar pregnancy. Int J Gynaecol Obstet 2015;131(3):281–4. https://doi.org/10.1016/j.ijgo.2015.06.029