

A rare case reporting history of CS scar ectopic managed expectantly ending in placenta previa then its recurrence in next pregnancy and managed by hysteroscopic resection: A case report and literature review

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SUMMARY

AUTHORS' CONTRIBUTION: (A) Study Design · (B) Data Collection · (C) Statistical Analysis · (D) Data Interpretation · (E) Manuscript Preparation · (F) Literature Search · (G) No Fund Collection

Introduction: A Cesarean scar ectopic pregnancy (CSEP) sometimes causes life-threatening bleeding and many treatment strategies have been proposed; expectant management, methotrexate administration, surgical evacuation, hysteroscopic resection, uterine artery embolization, or combination of them. We here report a patient: she had CSEP, which was managed expectantly and yielded placenta previa. Then, next, she had recurrent CSEP, which was resected hysteroscopically with success.

Case Presentation: The patient had abdominal pain and vaginal bleeding. Ultrasound and magnetic resonance imaging revealed the image-findings indicative of CSEP compatible with 5 weeks of pregnancy. Hysteroscopic resection of CSEP was performed with success.

Conclusion: CSEP can recur and physicians should be aware of this. Hysteroscopic resection may be an option of the treatment of CSEP.

Keywords: Case report; Recurrent; CS scar ectopic; Placenta previa; Hysteroscopic resection

List of abbreviations: CSEP: Cesaarean section ectopic pregnancy; CS: Cesaarean section; MRI: Magnetic Resonance Imaging; TVS: Transvaginal sonography; G: Gravida; P: Para

INTRODUCTION

A cesarean scar ectopic pregnancy (CSEP) is an implantation of pregnancy in the myometrium of lower uterine segment of a previous cesarean section scar. It is a rare condition and occurs in approximately one in every 2000 patients who have had a previous cesarean section [1]. Its incidence has increased with the increase in number cesarean deliveries. It carries a greater risk of maternal hemorrhage and ultimately maternal mortality. There are many modalities of options for treatment including expectant management, methotrexate, surgical evacuation or hysteroscopic resection, intervention radiological uterine artery embolization or a combined approach [2]. The level of success of each modality is dependent on the surgeon's skill and patient condition and choice. To the best of our knowledge that this is the first case to have a CSEP to be managed expectantly (and became a placenta previa in last pregnancy) and to recur in next pregnancy as CSEP.

CASE PRESENTATION

Patient specific information is not mentioned. She is G9P5+3 all deliveries by CS. The Primary concern of the patient is to prevent the occurrence of placenta accreta spectrum and its consequence of blood transfusion and peripartum hysterectomy. Obstetric history includes: 5 previous CS with last one was LSCS with midline laparotomy incision for placenta previa. Just before pregnancy was CSEP and end up in placenta previa. Physical examination (PE) revealed normal vital data, tenderness and rigidity all over the abdomen and marked cervical motion tenderness. Diagnostic testing done was BHCG was 7661miu/ml then repeated after 48 hours to be 18533 mIU/mL **Fig. 1.** TVS showed intrauterine gestational sac seen in the lower uterine segment at site of the C.S Scar with marked myometrial thinning. The gestational sac reveals smooth outline & show double wall sign with a yolk sac is seen within the gestation sac., but no fetal ploe is seen, GS 1.49 cm GA =5W 3D (picture suggestive of C.S scar ectopic pregnancy). MRI pelvis was done revealed: presence of a gestational sac within the endometrial cavity measuring 3.2 x 1.5x 2.1 cm bulging through the myometrium of the lower uterine segment in the region of Cesarean section scar. No diagnostic challenges were present this case. The diagnosis was confirmed as recurrent CSEP.

Hysteroscopic intervention was decided and the

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procedure began with cervical dilatation till Hegar dilator no 8, resectoscope was introduced revealing scar ectopic pregnancy with its lower pole attached to cesarean section

scar (**Fig. 2.**), resection of the lower pole was done (**Fig. 3.**) then suction curettage then insertion of intrauterine balloon tamponade (foley's catheter) filled with 50 ml

Fig. 1. Transvaginal ultrasound showing gestational sac at site of scar of CS.



Fig. 2. Hysteroscopy showing CS scar ectopic gestational sac

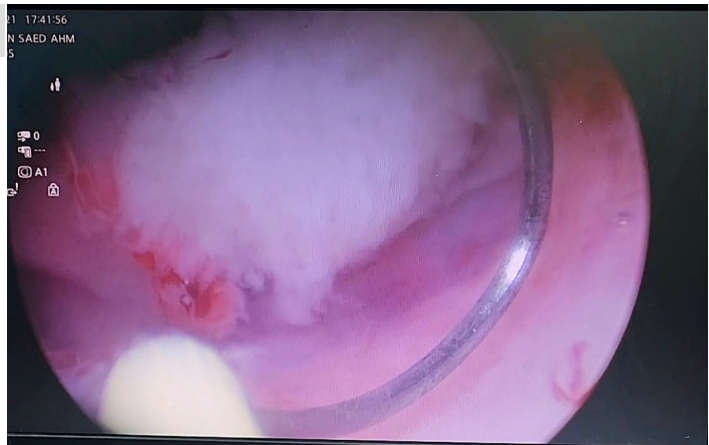
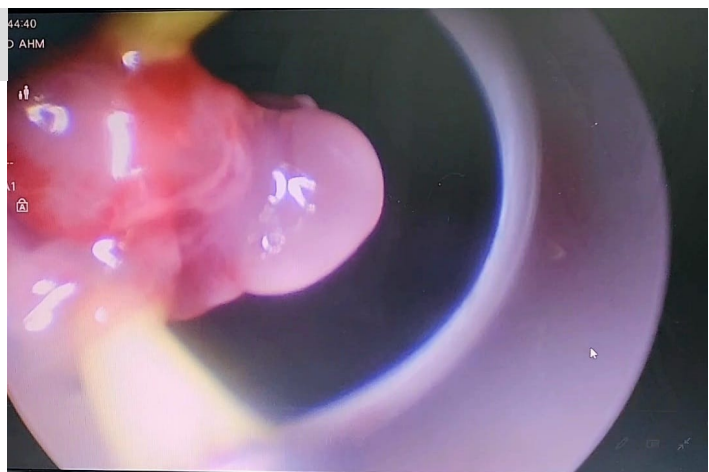


Fig. 3. Hysteroscopic removal of ectopic sac



saline. The procedure took around 60 minutes. The patient tolerated the procedure well with no Adverse and unanticipated events.

2nd day post-operative B-HCG was 3760, the intrauterine bleeding was minimal and the intrauterine catheter was removed after 48 hours. The patient was discharged after 72 hours.

DISCUSSION AND A LITERATURE REVIEW

The first case of a CSEP was recorded in medical field in 1978 in a 23 years old girl from South Africa [3]. As a result of the increasing numbers of caesarean sections of last decades, there has been increasing in the incidence of the pathology of these gestational abnormalities resulting in more physician orientation. The incidence of CSEP is reported in the literatures to be between 1:1800 to 1:2216 pregnancies with incidence of 0.15% in cases with previous caesarean sections, and the incidence is rising with increasing the number of repeated caesarean sections [4].

The pathophysiology of this condition remains unsure. A range of theories have been considered: (a) the gestational sac migrates endogenously through either a microscopic fistula within the cesarean section scar or a wedge defect in the LUS (lower uterine segment); (b) invasion of the uterine wall by placental villi wall at a site of scar dehiscence, and (c) implantation of the fertilized oocyte at the scar tissue areas of low oxygen tension [5]. This is explaining the pathology presenting in cases with previous procedures as cs (cesarean section), myomectomy, D&C (dilatation and curettage), hysterotomy, manual removal of placenta, and / or abnormal placentation in vitro fertilization as CSEP could be due to defects in the previous formed scar tissue, as a result of development of microtubular tract due to poor healing of the trauma caused by procedures [6].

The scar pregnancy has a different pathology in comparison to that of an intrauterine pregnancy with placenta accreta. In cases with placenta accreta formation, the conception's products are primarily present in the uterine cavity and the absence of decidua basalis is the main cause of varying degrees of invasion of the myometrium by trophoblastic tissues [7]. In cases of scar pregnancy, the myometrium and fibrotic tissues of the scar are completely surrounding the gestational sac, and separate it from the endometrial cavity [7]. The causing factor responsible for this pathology is the weak vascular support in the anterior uterine wall in some patients with history of previous caesarean section procedure, where blastocyst implantation occurs in the area of fibrous scar tissue generated by the previous CS (caesarean section) and to the myometrium before the formation of decidua basalis [8].

There are two types of scar pregnancy. Type I (endogenic form) is caused by implantation of gestational sac in the scar tissue of the previous caesarean section with growth towards the cervico-isthmic space or the uterine cavity [6,7]. In this type, a deep invasion in a caesarean scar tissue defect towards the urinary bladder and the abdominal

cavity is complicated by many adverse pregnancy outcomes such as ruptured uterus, intractable bleeding, emergency laparotomy and hysterectomy, and maternal death. Type II (exogenic form) refers to implantations growing inside the uterine cavity [6]. Type II is caused by deep implantation into scar defect tissues with infiltrating expansion into the uterine myometrium and uterine serosal surface, which may lead to ruptured uterus and severe hemorrhage in the first trimester of pregnancy, with a potential for loss of fertility, when massive hemorrhage ends by emergency laparotomy and hysterectomy [7].

Symptoms include first trimester vaginal bleeding and abdominal pain; but many women may be asymptomatic at diagnosis [9].

Transvaginal ultrasonography remains the golden standard modality in diagnosis CSEP in the first or early second trimester, which provides high resolution; however, it is recommended to combine color Doppler to grayscale evaluation, allowing more detailed visualization of the placental site implantation as well as definition of extraembryonic and fetal structures [10]. The Type I "on-the-scar" or endogenic form, mostly appears to have a considerable ultrasonographic clear layer of myometrium between the anterior wall of the uterus and the formed placenta. While the ultrasonographic pictures of Type II "in-the-niche" or exogenic form, show a thin myometrial interface below the placenta.

The diagnostic findings in the ultrasound suggestive of CSEP may also include: (1) an empty endocervical and endometrial cavity; (2), a nested gestational sac and placenta, on/in the area of previous scar; (3) the scar "niche" (the shallow are representing a healed hysterotomy site) is filled by a triangular ($\leq 8/40$ weeks), rounded or oval shaped gestational sac ($\geq 8/40$ weeks); (4) a thin (1–3 mm) or absent myometrial layer between the bladder and the gestational sac; (5) a unique or highly vascular pattern around the area of the scar, and (6) presence of an embryonic or fetal pole, yolk sac, or both with absence or presence of fetal cardiac activity. Bulging or ballooning of the lower uterine segment in the midline sagittal transabdominal view supports the diagnosis of CSEP [10,11]

In order to assure the maximum benefit from primary diagnosis and treatment, all pregnant women with an obstetric history of previous caesarean sections are advised to make first trimester scan at Early Pregnancy Assessment Clinic after a positive pregnancy test. transvaginal ultrasonography (TVS) remains the examination of choice, which might be combined with a transabdominal ultrasound in cases where a panoramic view is needed, and additional three-dimensional Power Doppler can confirm ultrasonographic findings. In equivocal cases, magnetic resonance imaging (MRI) may confirm the primary ultrasonographic diagnosis [12].

The available modalities of treatment are expectant management (an option which has been recently partially withdrawn by the recommendations of the Society for Maternal-Fetal Medicine), medical management

with methotrexate and surgical intervention; the case presentation and the clinical symptoms are the main factors in deciding the mode of treatment [10]. The available evidence in the literature favors surgical intervention rather than medical modality based on the success rates, although data are mainly based on case series, as summarized in the recent recommendations by the Society for Maternal-Fetal Medicine [10,13]. as clinical manifestations and potential complications are expected and might be expressed more seriously for CSEP. Therefore, surgical intervention remains the gold standard therapeutic procedure (combined with any other available approach).

Summarizing operative treatment options, besides the hysteroscopic (as in our case), laparoscopic or laparotomic surgical excision, vacuum aspiration and suction, can also be used to remove the scar pregnancy [10]. The current CSEP treatment modalities include medical management; medical management to be followed by uterine surgical treatment (usually minimally invasive), laparoscopic uterine incision and removal of the scar pregnancy foci, total laparoscopic hysterectomy (TLH), vaginal incision of the uterus and removal of CSEP elements with repair of the uterine muscle wall, in addition to selective uterine artery embolization (UAE) [14].

Management of CSEP in the 1st and early 2nd trimesters should be undertaken in a highly equipped center where a variety of treatment modalities and blood bank services are available. Immediate surgical intervention, (principally with a minimally invasive approach) should be done to all hemodynamically unstable patients. For hemodynamically stable patients, management options include either surgical or medical termination of pregnancy or even continuation of the pregnancy in special situations. In cases of CSEPs individuals with a fetal death, expectant management could be an option in combination with medical or surgical treatment [11,15]. Expectant management outcomes appear to be more favorable in patients with Type I rather than Type II CSEPs, especially in those where the myometrial thickness is ≥ 3 mm [16].

Operative resection of the CSEP can be performed hysteroscopically, laparoscopically, or with laparotomy [17]. Suction aspiration with guided ultrasound is an alternative CSEP approach in the early 1st trimester (5 to 7 weeks), with addition of a transcervical balloon catheter use in cases where massive bleeding takes place [17]. Transvaginal or transabdominal intragestational injection of methotrexate (MTX) under ultrasonographic guidance appears to be an effective treatment option for CSEP with 85% success rates in the early first trimester (6 to 8 weeks), however, in more advanced pregnancies it is difficult to predict the total effectiveness [19]. KCl injection Transabdominally or transvaginally under ultrasonographic guidance (5 mEq into the gestational sac) for a CSEP with fetal heart activity has also been described [18]. Medical treatment by systemic MTX can be administered as an adjunct to all of the above modalities [19].

CSEP should be treated without any delay once

diagnosis is confirmed, and a swift decision for termination should be made because of the increased risk for bleeding in case the pregnancy continuation [4]. The main principle of the treatment remains to terminate the pregnancy, removing the gestational sac, preserving the patient's fertility. Currently, most studies encourage that CSEP patients with severe type I or type II CSEP should receive UAE (uterine artery embolization) treatment, which is associated with marked decrease of the risk for developing intraoperative bleeding [14].

Only a few cases of laparoscopic management of CSEP have been reported. Recently, Kathopoulos et al. shared their experience with laparoscopic treatment of two cases utilizing different operative techniques. Laparoscopic management of CSEP, used either as a 1ry intervention or after failure of medical treatment, they consider removal of CSEP laparoscopically is mandatory when the scar pregnancy is growing towards the urinary bladder and abdominal cavity (type II CSEP) [20]. Laparoscopic excision of CSEP up to eleven weeks of gestation has also been recorded [21]. The main advantage of the laparoscopic approach is the complete removal of the retained products of conception at the time of the surgery leading to a less time for follow-up [4]. Moreover, restored uterine anatomy of the lower segment increases favorable future fertility outcomes [22]. Although representing a reliable management approach, it should be done by skilled laparoscopic surgeons.

The strength of this case report is that this is the first case to have a CSEP to be managed expectantly (and became a placenta previa in last pregnancy) and to recur in next pregnancy as CSEP. The weakness is that we do not have the investigations and MRI of previous CSEP that ended in placenta previa.

The message from this case report that we are reluctant to encourage surgeons about the expectant management of CSEP even if it is of endogenous type as its risk of placenta accretes or recurrence in next pregnancy remains high.

CONCLUSION

CSEP is a rare obstetrical condition, which may result in the woman to be in risk of a life-threatening situation such as ruptured uterus and massive hemorrhage, which may lead to maternal death. This situation represents a big diagnostic challenge in our obstetrics and gynecology clinical field, and management and careful decisions should be timely and made as soon as possible. Clinicians should depend on transvaginal ultrasonography as the primary diagnostic tool. Women should have access to all appropriate treatment options for CSP. Frequently, management should be individualized, as in our case, where decision for hysteroscopic resection treatment approaches was made in the concept of personalized medicine and the ideal management of such a life-threatening condition.

PATIENT PERSPECTIVE

Patient was satisfied with the procedure done as she is

convinced that expectant management done in previous CSEP would have endangered her life. The patient gave informed consent and it's available upon request.

DECLARATION

Ethics approval and consent to participate: An informed consent was taken from the patient to publish the case, No IRB is needed from our hospital in surgical case reports.

CONSENT FOR PUBLICATION

The patient gave the consent to publish case.

AVAILABILITY OF DATA AND MATERIALS

Available upon request.

COMPETING INTEREST

Authors declare no conflict of interest.

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