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Case Report

# Case Report Series: Ruptured Cesarean Scar and Ovarian Ectopic Pregnancy with Massive Hemoperitoneum

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## Abstract

An ectopic pregnancy is a pregnancy in which the developing blastocyst becomes implanted at a site other than the endometrium of uterine cavity. Most common extrauterine locations are the fallopian tube, however, incidence of implantation to other sites are rising and likely related to assistant fertility treatment and previous scar of uterus. Unfortunately, despite improvement in diagnosis, hemorrhage in first trimester of pregnancy is still a leading cause of maternal death.

Cesarean scar pregnancy is a rare, life threatening form of ectopic pregnancy where gestation sac partially or fully implanted to previous scar. If it progresses to second or third trimester, there would be an increased risk of uterine rupture, catastrophic bleeding and maternal death. Ovarian ectopic pregnancy also a rare event and not uncommon to be confused with hemorrhagic cyst or corpus luteal cyst during clinical evaluation and operation.

Emergency life threatening presentation of ectopic pregnancies are not uncommon, clinical judgement along with basic ultrasound skills can save the life of a woman. Despite working in high resource countries with all required facilities, emergency rare cases are still challenging. High risk behaviors, Immigrant status, insufficient health care access and unawareness of health care are a basic factor to expect increase in such presentation. Moreover, increased rate of caesarean section is a major risk factor for placental spectrum disease or scar pregnancy or scar niche.

We thereby, present a two interesting case reports of similar presentation of ovarian and scar pregnancy which, however share the same nature of disease but entirely different in location and etiology. Both presented in state of hemodynamic shock without proper early pregnancy assessment, both successfully managed by surgery and discharged in stable condition with appropriate follow up plan.

***Keywords:*** *Cesarean Scar; Ovarian Ectopic Pregnancy; Massive Hemoperitoneum*

## Introduction

Ectopic pregnancy incidence in UK is approximately 11/1000 pregnancies, with an estimated 11,000 ectopic pregnancies diagnosed each year [1]. The most common location is the fallopian tube, which accounts for 96 percent of all ectopic gestation. Ovarian pregnancy account for 3% and Cesarean scar pregnancy incidence is less than 1% (Figure 1). Prevalence of CSP estimated approximately 1 in 2000 of pregnancies (GTG). The risk factors include damaged fallopian tube secondary to infection, surgeries, use of uterine devices, smoking and *in vitro* fertilization [2].

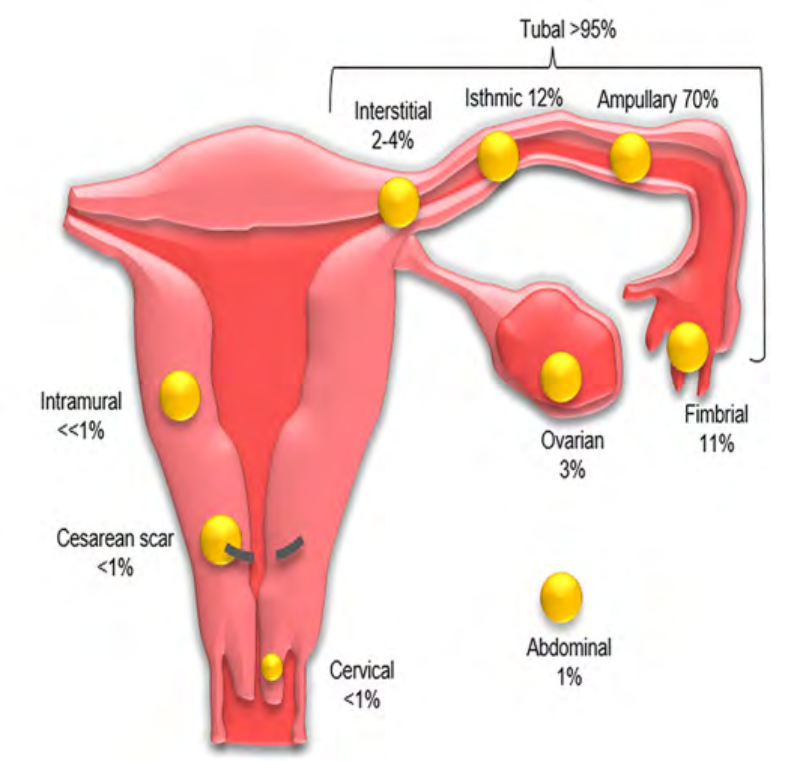


***Figure 1:*** *Schematic representation of ectopic pregnancies, location and frequencies (Source: ResearchGate, Springer Nature journal).*

Cesarean scar is associated with severe maternal morbidity and mortality. The fast and effective move of minimal invasive surgery and well researched medical treatment of ectopic pregnancy, there is dramatic changed in the management over the year. Preferred management is pharmacologic treatment with methotrexate [3,4] if maternal condition allows, size of mass is not very large and BHCG is not very high. Irrespective of location of pregnancy, it is important to remember that, it is still the leading cause of maternal death in first trimester of pregnancy and accounts for 4 percent of all pregnancy-related deaths [5]. It is worth noting to all health care professional working in gynecology or emergency set up that any young or reproductive age group woman attending to hospital with typical or atypical presentation of abdominal pain, ectopic pregnancy should be a top differential diagnosis requiring urgent work up before any deterioration. Despite improved diagnostic methods leading to earlier detection and treatment, unfortunately, ectopic is a leading cause of maternal death in first trimester. There is an extensive research and robust evidences regarding the best possible treatment modalities and options if patient attends the hospital in stable condition and time factor is in favor of medical or conservative or planned surgical approach. Unfortunately, many of ectopic pregnancies still present in state of hemodynamic instability and requiring urgent life saving surgical approach. Our cases are one of the examples of best, fast, effective management relative to sick women attending emergency without any prior investigations or ultrasounds.

Clinicians should be aware that transvaginal ultrasound is the primary diagnostic modality [3]. Clinicians should have an appropriate training or privilege to diagnose the ectopic pregnancy by clinical judgement supplemented by ultrasound. They should have a basic skill to perform emergency ultrasound to exclude intrauterine pregnancy or identify hemoperitoneum, this will help aiding the counselling and avoid litigations of surgical intervention or methotrexate use.

## Case Reports

Our first interesting case is about 31 years old, Asian woman, Para 2+4, one vaginal delivery followed by Cesarean section 2 years ago due to twin. Then she had 3 consecutive first-trimester spontaneous miscarriages, two managed by surgical evacuation. The last pregnancy was complicated with tubal abortion which managed expectantly until beta HCG dropped to 0. She brought by ambulance with history of severe lower abdominal pain at 7+ weeks of gestation associated with fainting episode. Unfortunately, there was no proper antenatal booking in this pregnancy despite having multiple risk factors. Reportedly having one ultrasound in private which suggested a missed miscarriage- report was not seen on arrival. On arrival to emergency, she was in state of hemodynamic shock, conscious but not well-oriented. she was very pale with cold extremities. Blood pressure was 80/50 mm/Hg, heart rate was 120 beats per minute, saturation was 95 percent with 10L oxygen. Abdominal examination reveals generalized abdominal tenderness with rigidity and distended abdomen. Bimanual digital vaginal examination suggested a small uterus with fullness all around the uterus. Transabdominal and transvaginal ultrasound suggested a distorted uterine anatomy. intrauterine gestational sac at lower uterine part. moderate free fluid with lot of clots all over the pelvic cavity. After initial resuscitation, exploratory laparotomy was performed.

Operative findings consistent with enlarged Bicornuate uterus with bilateral tubes and ovaries normal. Peritoneal cavity full of clots and fresh blood around 2000 cc.

Active bleeding seen at right lower uterine part near the previous scar close to bladder. On dissection of adherent bladder, trophoblastic tissues seen protruding through the rupture scar. tissues removed and bladder separated by urologist.

Rent on uterus extended as previous scar found deficient and thin and right end of it had given way. Cavity further explored. Bicornuate uterus noted externally and septum felt internally at fundus. Cavity found empty. she received massive blood transfusion during and after operation and stayed in ICU for 2 days. Her initial hemoglobin was 6.9g per dL. After massive blood transfusion of packed cells and cryoprecipitate, she discharged in a stable condition with hemoglobin 10.7 gm/dl. Histopathology showed partial molar pregnancy with BHCG, 23,450 mIU/L at time of admission. She was regularly followed in clinic with serial BHCG which progressively dropped to less than 1 in 8 weeks.

Second case presented with similar symptoms. She was 22 years old woman of Asian ethnicity, nullipara, new in relation, attended emergency department with complaints of 6 weeks amenorrhea with positive pregnancy test at home. Presenting complaints was severe lower abdominal pain for few hours, pain was progressive in nature, moderate to severe in intensity, aggravated with change in position and associated with spotting. She declined any previous vaginal infections, any abdominal surgery or use of contraception- she was non-smoker. Body mass index was 33 kilogram/meter sq.

Although she was conscious and oriented but look very pale and hypotensive and tachycardic. Blood pressure was low 90/50 mm/Hg, heart rate was high 115/minutes, respiratory rate and oxygen saturation was normal.

Abdominal examination revealed generalized tenderness with guarding, rigidity and rebound. Speculum examination showed healthy cervix with minimal brownish discharge. Digital vaginal examination reflected a bulky, tender uterus with severe cervical excitation. Clinical findings were suggestive of ruptured ectopic with hemodynamic state of shock with compensatory phase.

Transabdominal ultrasound with vaginal ultrasound was performed in emergency which was suggestive of absence of intrauterine gestational sac, endometrial thickness was 8 mm, there was no clear adnexal mass was visualized except left ovarian cyst around 2.0 x 2.9 cm- there was gross hemoperitoneum with clots, Also, uterus was floating in the fluid- patient was immediately resuscitated with standard protocol and prepared for emergency laparoscopy after explaining the procedure and obtaining the consent- later she required inotropic

support to maintain the blood pressure. Palmer point entry was done, pelvic cavity was found full of blood clots and active bleeding. Uterus and both tubes and right ovary were healthy and normal. Left ovary was enlarged with suspected bleeding. Laparoscopy converted to laparotomy due to limited view in presence of massive hemoperitoneum and difficulty in identifying the source. The left ovarian corpus luteal like cyst was seen bleeding from ruptured site with suspected trophoblastic tissues. Abnormal tissues were removed, small biopsy was taken and the capsule sutured, tissue sent for histopathology. There was gross hemoperitoneum and estimated blood loss was 2000 cc. She received 3 units of packed cells during operation and 2 thereafter. Histopathology was Consistent with ovarian corpus luteum and ectopic gestation.

Her initial beta HCG at time of admission was 5,332.00. Beta HCG after surgical management and after 48 hours was 1,946.00 and 607.00 respectively. Initial hemoglobin was 9.1 which could be misleading due to severe hemorrhage and hemoconcentration. After receiving massive transfusion, she built up her hemoglobin 11.1 grams/deciliter. She discharged in stable condition on second day of surgery with advice of weekly beta-hCG till it become negative. Also, was advised against pregnancy and barrier methods of contraception was suggested.

## Discussion

By successful management of rare cases of scar and ovarian pregnancies, we can confidently conclude that despite working in high income countries with available resources, maternal collapse in early pregnancy related to ectopic is not uncommon. Scar ectopic are rising considering previous uterine surgeries, however, young woman with first pregnancy without risk factors can present with critical state of shock. Travelling and relocation is constantly rising globally. Inappropriate immigrant status, financial constraint and lack of awareness of healthcare facilities are the main risk factor for categorizing the pregnancy in high-risk group from the beginning of pregnancy.

Cesarean scar pregnancy: If we look at the first case of CSP, we see the combination of high-risk factors including, previous scar, previous ectopic pregnancy, previous two surgical treatment of spontaneous miscarriages, consecutive recurrent miscarriages and the woman belongs to vulnerable class of low socio-economic group, not attended the health care system early. UK and Ireland confidential inquiries of maternal death and maternal morbidities December 2022, states that the maternal mortality rate amongst women who live in the most deprived areas is increasing and addressing these disparities must remain an important focus. Vulnerable and young women remain disproportionately represented among those who died from ectopic pregnancy. They need additional safety measures incorporated into their care, for example, enhanced follow-up pathways. Each contact with girls or women of childbearing age following miscarriage, prescribing contraception, at sexually transmitted infection screening and at smear tests is an opportunity to educate regarding red flag symptoms associated with ectopic pregnancy. The awareness of symptoms may reduce deaths amongst vulnerable women and teenage girls.

Caesarean scar pregnancy (CSP) is a rare form of ectopic pregnancy whereby the gestational sac is fully or partially implanted within the scar caused by a previous caesarean section (CS). This is attributed partly to the increasing number of CS performed and also to increasing awareness and better ultrasound diagnosis. The first case was reported in 1978 [6]. Estimates of CSP incidence range from 1/1800 to 1/2500 of all pregnancies, less than 1%. It has been estimated that 6.1% of pregnancies in women with at least one previous CS will have a diagnosis of cesarean scar ectopic pregnancy [7-9].

### Pathophysiology

Little is known about the mechanism and an etiopathology of CSP. Endometrial and myometrial disruption or scarring could be predisposing factors in abnormal pregnancy implantation. The most probable mechanism explaining scar implantation is invasion by the implanting blastocyst through a microscopic tract that develops from the trauma of an earlier CS or other uterine surgeries [10]. The presence of a CS scar in the uterus may also inhibit implantation of the gestational sac secondary to the more global effect of prior surgery

on the endometrium, rather than just the physical presence of a scar. The risk of scar implantation might be proportional to the size of the anterior uterine wall defect, possibly caused by a larger surface area induced by the scar [11,12].

### Diagnostic criteria and treatment modalities CSP

Transvaginal ultrasound is a gold standard test for diagnosing CSP. BHCG can help to facilitate decision modalities, however it is not included in diagnostic criteria. MRI considered as second line and both share essentially same features. Transvaginal ultrasound is superior considering availabilities in emergency and cost effective.

The diagnostic criteria described for diagnosing caesarean scar implantation on transvaginal ultrasound include:

1. Empty uterine cavity
2. Gestational sac or solid mass of trophoblast located anteriorly at the level of the internal os embedded at the site of the previous lower uterine segment caesarean section scar.
3. Thin or absent layer of myometrium between the gestational sac and the bladder.
4. Evidence of prominent trophoblastic/placental circulation on Doppler examination.
5. Empty endocervical canal [13].

### Treatment modalities

As we discussed, these pregnancies may be ongoing potentially viable pregnancies or miscarriages within the scar. Medical and surgical interventions with or without additional hemostatic measures should be considered in women with first trimester caesarean scar pregnancy [Green top guideline-21]. There is insufficient evidence to recommend any one specific intervention over another for caesarean scar pregnancy, but the current literature supports a surgical rather than medical approach as the most effective (Green top guideline 21). Methotrexate is drug of choice for medical management and commonly used systematically. Local embryocides injection have also been used including Methotrexate, potassium chloride and etoposide and hyperosmolar glucose [14]. Combined local and systemic Methotrexate has also been used. Surgical management include dilatation and curettage, hysteroscopic resection, abdominal or laparoscopic resection. They can be used as primary treatment or sequential treatment after methotrexate or uterine artery embolization.

There is possibility of misdiagnosis of scar pregnancies in early pregnancy and literatures says that thirteen percent of reported cases of caesarean scar pregnancy were misdiagnosed as intrauterine or cervical pregnancies at presentation [15]. The true prevalence of caesarean scar pregnancies is likely to be somewhat higher than estimated in the literature as some cases will end in the first trimester, either by miscarriage or termination, and go unreported and undiagnosed. There is a spectrum of severity associated with pregnancies implanted into caesarean section scars and the natural history is uncertain. Vial., *et al*. [16] proposed that there are two different types of pregnancies implanted in a caesarean scar: the first type progressing into the uterine cavity as the gestational sac grows and develops, so with the potential to reach a viable gestational age, but with the risk of massive bleeding from the implantation site; and the second with progression deeper towards the serosal surface of the uterus with the risk of first trimester rupture and hemorrhage. This explanation supports our case of CSP rupture in first trimester and presented in state of shock. With Given that there are also varying appearances of caesarean section scars on the uterus and that placental development evolves over time as the pregnancy progresses, all of these factors can cause difficulty in the diagnosis of caesarean scar pregnancy.

While retrospective analysis of our case, it seems that the intrauterine gestational sac was implanted deep towards the serosal surface and subsequently misdiagnosed as spontaneous miscarriage requiring follow up- Patient missed the further care and attended in emergency with catastrophic bleeding and life-threatening shock.

There have also been numerous small case series and case reports 42 of intra-abdominal rupture and severe vaginal hemorrhage at the point of diagnosis or following intervention. There were six maternal deaths due to hemorrhage in women with a history of caesarean section in the 2006 - 2008 Centre for Maternal and Child Enquiries report (CMACE) [17] although, the site of implantation was not always identified.

All treatment options carry a risk of hemorrhage and subsequent hysterectomy. Treatment should be individualized based on full pre-treatment evaluation. In principle, pregnancy should be ended as soon as possible after confirming the diagnosis, with the aim of removing the gestational sac and the CSP mass to retain future fertility.

### Ovarian pregnancy

Ovarian pregnancy is a rarest form of extrauterine pregnancy and refers to a pregnancy that is located in the ovary. Typically, the egg cell is not released or picked up at ovulation, but fertilized within the ovary where the pregnancy implants. Such a pregnancy usually does not proceed past the first four weeks of pregnancy; However, it may go beyond as our case presented at 6 weeks plus weeks gestational amenorrhea. The cause of primary ovarian pregnancy remains unknown and it would seem to be secondary to reflux of the fertilized oocyte to the ovary [18]. The cases of ovarian pregnancy after IVF reported in the literature support the theory of reflux [19]. Other hypotheses have suggested interference in the release of the ovum from the ruptured follicle, malfunction of the tubes and inflammatory thickening of the tunica albuginea. Hertig estimated that ovarian pregnancy occurs in one in 25,000 to 40,000 pregnancies, comprising approximately 3% of ectopic pregnancies [20].

Apart from above explanation rest of the etiology shares the same pathophysiology of other ectopic pregnancies including infection and inflammatory response surrounding the tubal area. Its presentation often is similar to other tubal ectopic pregnancies and difficult to distinguish from that of tubal ectopic pregnancy and hemorrhagic ovarian cyst.

Green top 2016 states that there are no specific agreed criteria for the ultrasound diagnosis of ovarian ectopic pregnancy. A baseline serum BHCG should be carried out at diagnosis to help with diagnosis. In some cases, 48 hours repeat BHCG may be required for further management.

In diagnosed and stable cases systemic Methotrexate can be used to treat ovarian ectopic, however, minimal access surgery is now the treatment of choice for ovarian ectopic pregnancy. Removal of the gestational products by enucleation or wedge resection (in the presence of a large ectopic mass) is preferred. Enucleating the products of conception bluntly from the ovary minimizes damage to the surrounding ovarian tissue. Hemostasis can be achieved by electrocautery or suturing. Oophorectomy is occasionally required when there is coexisting ipsilateral ovarian pathology or excessive bleeding [21].

Ovarian pregnancies can be confused for hemorrhagic or corpus luteal cysts even at laparoscopy. Referring to our case of a nullipara, intraoperative findings at laparoscopy suggested a healthy bilateral normal tube, normal uterus with fresh bleeding and lot of clots in abdominal cavity which were obscuring the view and interfering with identifying the source and controlling hemostasis. Laparoscopy converted to laparotomy and active bleeding found on left ovary. It seems a ruptured corpus luteal cyst or an ectopic pregnancy, subsequently diagnosed ovarian ectopic pregnancy on histopathology. Initial BHCG was 5332 Miu/L which gradually dropped to zero.

## Conclusion

Emergency life threatening presentation of ectopic pregnancies are not uncommon, clinical judgement along with basic ultrasound skills can save the life of a woman. Despite working in high resource countries with all available resources, emergency rare cases are still challenging. High risk behaviors, Immigrant status, insufficient health care access and unawareness of health care are a basic factor to

expect increase in such presentation. Moreover, increased rate of caesarean section is a major risk factor for placental spectrum disease or scar pregnancy or scar niche. Cesarean section or planned birth has become a fashion or trend in last few decades. Here, the health care professional should pause and review the maternal request for caesarean delivery. Counselling plays an important role for choosing a mode of delivery. Appropriate information regarding future pregnancy and delivery should be clearly informed to woman which may include but not limited to serious future morbidities: scar pregnancy, placenta previa spectrum disorders, rupture uterus, horrendous bleeding and ICU stay. Woman request should be explored empathically. Appropriate analgesia, support and avoidance of previous traumatic environment help encouraging woman for making informed choice of delivery. Mental health assessment may require if no valid reason is expressed by woman.

Despite having robust evidences of various modalities of managing scar and ovarian ectopic pregnancy, secret of life saving is fast decision to proceed to surgery with skilled and experienced surgeon.

## Conflict of Interest

The authors have declared that no competing interests exist.

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