

## CASE REPORT

# Ectopic Pregnancy in Isthmocele: A Report of Unrecognized Case

<sup>1</sup>Nutan Jain, <sup>2</sup>Swati Varshney

## ABSTRACT

Cesarean scar pregnancy (CSP) is one of the rarest forms of ectopic pregnancy. Little is known about its incidence and natural history. The diagnosis and treatment of CSP is challenging. The authors report a case of CSP with heavy bleeding per vagina, who underwent suction and evacuation after abortion; patient underwent hysterolaparoscopy with resection of ectopic mass. The patient was discharged from the hospital without any complications. The CSP should be diagnosed and treated as soon as possible to prevent severe complications and spare fertility.

**Keywords:** Cesarean scar defect, Ectopic pregnancy, Isthmocele.

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## INTRODUCTION

According to the latest global estimates, approximately 15% of deliveries take place by cesarean section (CS), with about 20 million CS deliveries occurring each year worldwide.<sup>1</sup> Such a proportion has steadily increased in almost all middle- and high-income countries over the last three decades. Several obstetric complications due to inappropriately healed cesarean scar, such as placenta accreta, scar dehiscence, and scar ectopic pregnancy are increasingly reported along with rising cesarean rates.

Women may incur a poor uterine scar healing after CS, which can cause anatomic defects of the uterine cavity of various degrees.<sup>2</sup> Such defects may lead to gynecologic sequelae, such as abnormal bleeding, pelvic pain, infertility, and most feared complications of cesarean scar pregnancy (CSP) and uterine rupture during subsequent pregnancy and labor.<sup>3</sup> Such anatomic defects can have

different shapes, such as thin linear defect, focal saccular outpouching, unilateral or bilateral diverticula, and fistula at different locations, such as the uterine body, lower uterine segment, uterine isthmus, and the upper endocervical canal.<sup>4</sup>

Such defects are often termed as isthmocele/previous cesarean scar defect (CSD)/uterine niche, defined as diverticulum at lower uterine segment cavity, uterine isthmus, or endocervical canal at the site of previous CS scar. In CSP, the gestational sac is implanted in the myometrium at the site of a previous CS.

Implantation of a pregnancy within a cesarean fibrous tissue scar is considered to be the rarest form of ectopic pregnancy and a life-threatening condition.<sup>5</sup> This is because of the very high risk for uterine rupture and all the maternal complications related to it. Of the many theories for explaining its occurrence, the most reasonable one seems to be that the blastocyst enters into the myometrium through a microscopic dehiscence tract. This may be created through a trauma of a previous CS, any other uterine surgery, or even following manual removal of the placenta.<sup>5</sup>

We present our medical center's experience of ectopic pregnancy implanted in cesarean scar.

## CASE REPORT

A 31-year-old female came to the Department of Obstetrics and Gynaecology in our hospital with complaints of heavy bleeding per vagina since 3 months and continuous bleeding per vagina since 10 days. Obstetric history of patient revealed that she had undergone CS one and half years back in 2015. In October 2016, there is history of being overdue by 10 days followed by bleeding per vagina. Urine pregnancy test done at that time was positive, and suction and evacuation were done at that time. After that, patient was having regular bleeding every month, but flow was heavy, and since last 10 days, bleeding was continuous and heavy.

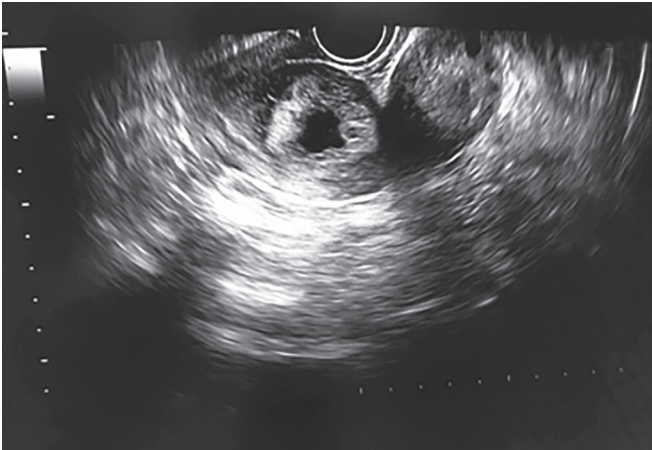
Transvaginal scan done outside showed a well-defined heterogeneous lesion of 35 × 40 mm with areas of central cystic component in cervix with predominant involvement of anterior wall projecting into endocervical canal suggestive of degenerating fibroid.

Complete examination and evaluation were done in our hospital. Transvaginal scan was done, which showed

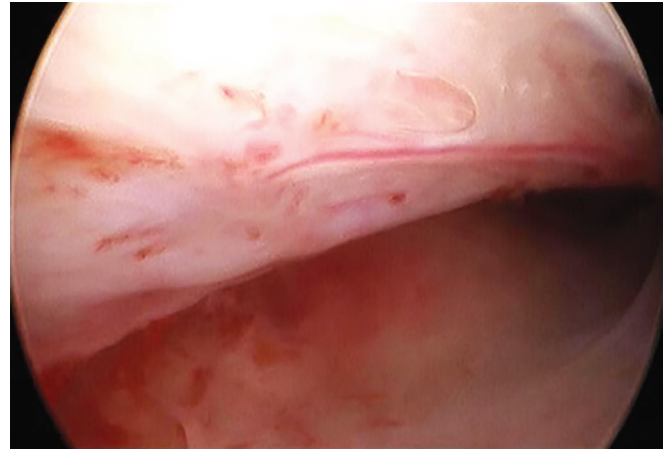
<sup>1</sup>Director, <sup>2</sup>Clinical Research Fellow

<sup>1,2</sup>Department of Gynaecology and Obstetrics, Vardhman Trauma & Laparoscopy Centre, Muzaffarnagar, Uttar Pradesh India

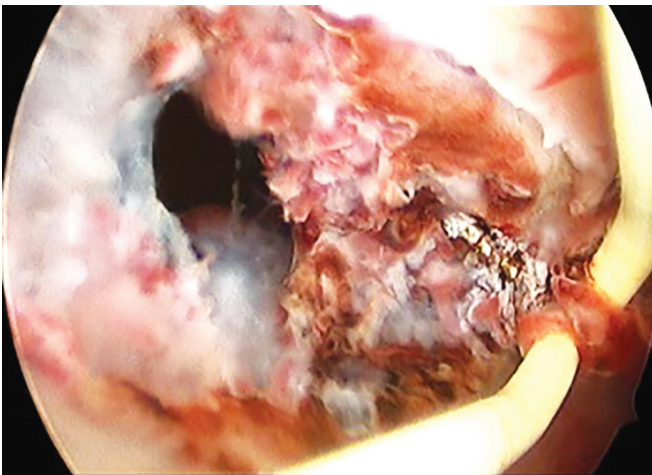
**Corresponding Author:** Nutan Jain, Director, Department of Gynaecology and Obstetrics, Vardhman Trauma & Laparoscopy Centre, Muzaffarnagar, Uttar Pradesh, India Phone: +911312623084, e-mail: jainnutan@gmail.com



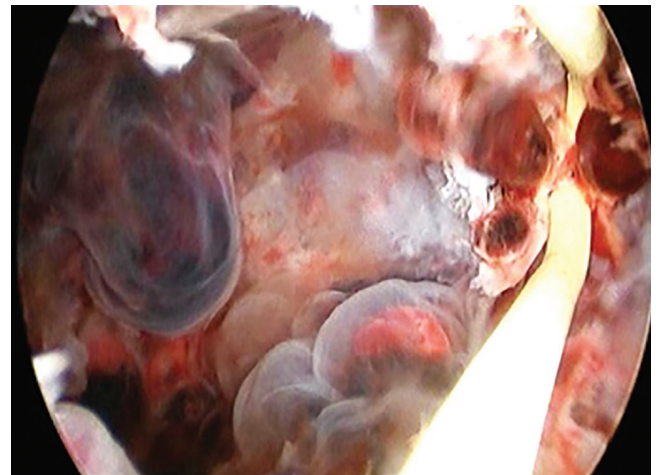
**Fig. 1:** Ultrasound picture showing ectopic pregnancy in isthmocele



**Fig. 2:** Trophoblastic villi were seen as intense white fronds



**Fig. 3:** Incision made at right angle to crescent



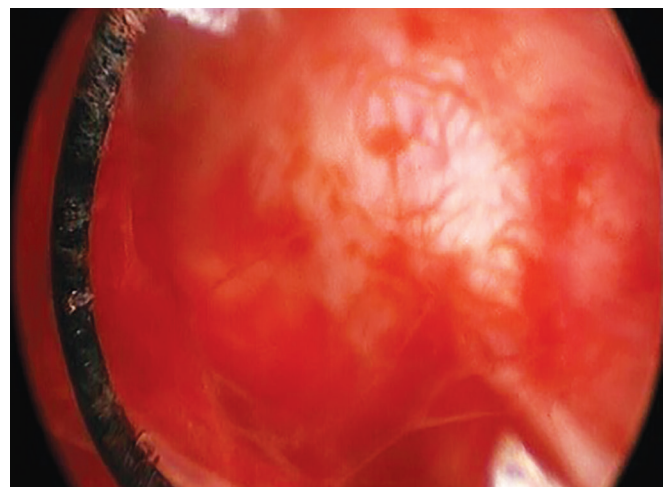
**Fig. 4:** Bluish purple products of conception

scar defect in lower uterine segment and  $40 \times 32$  mm mass near scar defect suggestive of ectopic pregnancy (Fig. 1).

After proper examination, investigation, and anesthesia fitness, patient was taken up for hysterolaparoscopy evaluation.

On laparoscopy, dense omental adhesions were noted on the anterior abdominal wall. All adhesiolysis done, uterus was visualized to appear normal in size, shape, and position.

Hysteroscopy revealed normal uterine cavity with a crescent-shaped bulge in lower uterine cavity up to upper cervical canal (Fig. 2). Incision was made by resectoscope using Collins knife (Fig. 3). After making an incision exactly at right angles to the crescent, bluish purplish products of conception were noted (Fig. 4). An intact gestation sac also came into view. All these products were removed using resectoscope. After removal of products, a clear diverticula-like cavity was noted, but now it was communicating with the uterine cavity above and cervical canal below (Fig. 5).



**Fig. 5:** Normal upper uterine cavity

## DISCUSSION

As the incidence of CS is on the increase worldwide, the complications associated with them are becoming more common. Therefore, the effect of postcesarean complications on women's health is a focus of increasing attention.



The CSP is the rarest kind of ectopic pregnancy, but because of the increasing number of cesarean deliveries, its incidence has been rising to be about 1/2,000 normal pregnancy.<sup>6</sup> The incidence does not appear to correlate with the number of cesarean deliveries. The mechanism for implantation in this location is believed to be migration of the embryo through either the wedge defect in the lower uterine segment or a microscopic fistula within the scar.<sup>7,8</sup>

Many of these operations are currently elective procedures performed in a nondeveloped lower uterine segment, so that the healing processes following the operations might facilitate implantation of the blastocyst within the scar. Multiple (>2) CSs seem to be another risk factor for in-scar implantation of the subsequent pregnancy because of increased scar surface area.<sup>9</sup> In addition, the increasing number of CSs currently performed, together with the changing surgical technique might also have the same impact. In the past, the uterus was closed using a double layer of multiple sutures inverting the first layer with the second row. However, a single-locking suture technique is currently more frequently used.

Most of the cases that have been reported were diagnosed early in the first trimester. Differential diagnosis includes spontaneous abortion in progress and cervical ectopic pregnancy. Diagnosis is difficult, but often possible using vaginal ultrasonography and color flow Doppler.<sup>1</sup> Proposed ultrasound diagnostic criteria, allowing a differential diagnosis with cervical ectopic pregnancy, are (i) a gestational sac located between the bladder wall and the anterior isthmic portion of the uterus; (ii) no trophoblastic tissue visible in the uterine cavity and cervical canal; (iii) clearly visible circular blood flow surrounding the sac; (iv) a thin myometrial layer between the bladder and gestational sac; and (v) a triangular-shaped gestational sac.<sup>10-12</sup> When the pregnancy is not localized by ultrasonography, either laparoscopy or hysteroscopy can be used for the diagnosis.

Here, we discussed a case of ectopic pregnancy in a woman having a history of CS one and half year back, and history of urine pregnancy test positive 4 months back followed by bleeding per vagina, who complained of heavy bleeding per vagina since abortion and continuous bleeding per vagina since 10 days. In this case, the ectopic pregnancy in isthmocele was unrecognized at sonography, due to its unusual location within the anterior wall of the uterus in lower segment, misinterpreted previously as the degenerating fibroid.

A widely accepted approach for the treatment of CSD ectopic pregnancy has not been developed. Systemic methotrexate therapy, gestational sac aspiration, dilatation and curettage, surgical laparotomy/hysterotomy, hysteroscopy, laparoscopy, and uterine artery embolization are

used. Ultimately, the approach depends on various factors, such as gestational age at presentation, hemodynamic stability, local endoscopic expertise, future fertility plans, and feasibility of serial follow-up serology and imaging. Hysteroscopy is the "gold standard" for CSD assessment; in fact, it enables confirming diagnosis and assessing treatment.<sup>7,13</sup> Hysteroscopic surgical procedures are aimed at resecting the fibrotic tissue that sometimes hangs below the scar, thereby improving menstrual drainage and avoiding blood accumulation. Furthermore, hysteroscopy enables the fulguration of superficial dilated blood vessels or endometrium-like glands inside the diverticulum to avoid the in situ production of fluid or blood.

In a recent review, Tower and Frishman<sup>3</sup> observed that CSD and isthmocele are underrecognized causes of abnormal uterine bleeding and other gynecologic complications, and this case confirms such remark.

A recent systematic review by Roberge et al<sup>2</sup> showed that hystero-graphy, sonohystero-graphy (SHG), or transvaginal ultrasound can all detect uterine scar defects, with SHG having higher detection power. With ultrasound, the thickness of the residual myometrium, the thickness of myometrium bordering the scar, and the depth of the filling defect in the scar can all be depicted.<sup>14</sup>

Although exceptional, this case highlights the possibility that an ectopic pregnancy in isthmocele could be misinterpreted as degenerating myoma. In this case, combined approach of laparoscopy and hysteroscopy confirms the diagnosis and treatment.

## CONCLUSION

Ectopic pregnancy within the scar of a previous cesarean delivery can lead to uterine rupture and life-threatening intraperitoneal hemorrhage during the first trimester of pregnancy.

Though a rare event, the incidence of CSP seems to be on the rise. An obstetrician is likely to encounter this entity in his or her lifetime. In women with a history of CSP, early ultrasound should be performed in subsequent pregnancies in order to establish the location of implantation.

## REFERENCES

1. Betran AP, Merialdi M, Lauer JA, Bing-Shun W, Thomas J, Van LP. Rates of caesarean section: analysis of global, regional and national estimates. *Paediatr Perinat Epidemiol* 2007 Mar;21(2):98-113.
2. Roberge S, Boutin A, Chaillet N, Moore L, Jastrow N, Demers S, Bujold E. Systematic review of cesarean scar assessment in the nonpregnant state: imaging techniques and uterine scar defect. *Am J Perinatol* 2012 Jun;29(6):465-471.
3. Tower AM, Frishman GN. Cesarean scar defects: an under-recognized cause of abnormal uterine bleeding and other gynecologic complications. *J Minim Invasive Gynecol* 2013 Sep-Oct;20(5):562-572.

4. Ahmadi F, Torbati L, Akhbari F, Shahrzad G. Appearance of uterine scar due to previous cesarean section on hysterosalpingography: various shapes, locations and sizes. *Iran J Radiol* 2013 Jun;10(2):103-110.
5. Fylstra DL, Pound-Chang T, Miller MG, Cooper A, Miller KM. Ectopic pregnancy within a caesarian delivery scar: a case report. *Am J Obstet Gynecol* 2002 Aug;187(2):302-304.
6. Rotas MA, Haberman S, Levгур M. Cesarean scar ectopic pregnancies: etiology, diagnosis, and management. *Obstet Gynecol* 2006 Jun;107(6):1373-1381.
7. Gubbini G, Casadio P, Marra E. Resectoscopic correction of the "isthmocele" in women with postmenstrual abnormal uterine bleeding and secondary infertility. *J Minim Invasive Gynecol* 2008 Mar-Apr;15(2):172-175.
8. Marchiolé P, Gorlero F, de Caro G, Podestà M, Valenzano M. Intramural pregnancy embedded in a previous cesarean section scar treated conservatively. *Ultrasound Obstet Gynecol* 2004 Mar;23(3):307-309.
9. Jurkovic D, Hillaby K, Woelfer B, Lawrence A, Salim R, Elson CJ. First-trimester diagnosis and management of pregnancies implanted into the lower uterine segment caesarian section scar. *Ultrasound Obstet Gynecol* 2003 Mar;21(3):220-227.
10. Sugawara J, Senoo M, Chisaka H, Yaegashi N, Okamura K. Successful conservative treatment of a cesarean scar pregnancy with uterine artery embolization. *Tohoku J Exp Med* 2005 Jul;206(3):261-265.
11. Vial Y, Petignat P, Hohlfeld P. Pregnancy in a cesarean scar. *Ultrasound Obstet Gynecol* 2000 Nov;16(6):592-593.
12. Seow KM, Hwang JL, Tsai YL. Ultrasound diagnosis of a pregnancy in a cesarean section scar. *Ultrasound Obstet Gynecol* 2001 Nov;18(5):547-549.
13. Fernandez E, Fernandez C, Fabres C, Alam VV. Hysteroscopic correction of cesarean section scars in women with abnormal uterine bleeding. *J Am Assoc Gynecol Laparosc* 1996 Aug;3 (Suppl 4):S13.
14. Regnard C, Nosbusch M, Fellemans C, Benali N, van Ryselberghe M, Barlow P, Rozenberg S. Cesarean section scar evaluation by saline contrast sonohysterography. *Ultrasound Obstet Gynecol* 2004 Mar;23(3):289.