

CASE REPORT

Pregnancy in an isthmocele: A rare case from Nepal

Alina Tandukar¹ | Roshan Aryal²  | Bishal Khaniya¹ | Suvana Maskey¹ | Neebha Ojha¹ | Dinesh Chataut³

¹Department of Obstetrics and Gynecology, Tribhuvan University Teaching Hospital, Kathmandu, Nepal

²Maharajgunj Medical Campus, Institute of Medicine, Kathmandu, Nepal

³Department of Radiology and Imaging, Tribhuvan University Teaching Hospital, Kathmandu, Nepal

Correspondence

Roshan Aryal, Maharajgunj Medical Campus, Institute of Medicine, Maharajgunj, Kathmandu, Nepal.
Email: roshanaryal7@gmail.com

Abstract

An infrequent form of ectopic pregnancy, pregnancy in an isthmocele can be hazardous due to hemorrhage or uterine rupture. With no clear guidelines for the management of this condition, surgery is the preferred option.

KEYWORDS

cesarean scar defects, ectopic pregnancy, isthmocele

1 | INTRODUCTION

Isthmoceles, also known as scar defects at the isthmus uteri or cesarean scar defects, are found to be associated with spate of gynecological and obstetrical issues, often after cesarean delivery.¹⁻⁴ With the incidence ranging from 6.9% to 69%, the presence of an isthmocele is associated with morbidly adherent placenta, cesarean scar ectopic pregnancy, or uterine rupture.^{5,6}

Here, we report a rare case of ectopic pregnancy in an isthmocele in a 23-year-old woman.

2 | CASE REPORT

A 23-year-old P1 A1 woman presented with irregular per vaginal (PV) bleeding for 2 months. She underwent emergency lower section cesarean section (LSCS) 20 months back for fetal distress and resumed her regular menstrual cycle after 6 months of delivery, but at 17 months post-delivery she had heavy menstrual bleeding associated

with dizziness. Patient underwent dilatation and evacuation twice and even required blood transfusion before visiting our center.

On clinical examination, the hemodynamic state was stable and abdominal palpation was unremarkable. Per-speculum examination showed no bleeding and bimanual examination revealed normal sized, retroverted, and mobile uterus without any lateral mass.

Urine pregnancy test was positive and serum beta-human chorionic gonadotropin level was 55.06 $\mu\text{mol/L}$. Transvaginal ultrasound showed a defect at the region of cesarean incision site being replaced by cystic lesion in anterior and right side with no vascularity noted within, with differential diagnosis of an isthmocele as shown in [Figure 1](#). Magnetic resonance imaging (MRI) revealed 3.2 \times 2.4 \times 2.9 cm non-enhancing lesion in anterior myometrium with extension to the endometrial cavity through a focal defect in the lower uterine segment with the intact serosa, features likely of contained rupture at previous cesarean scar site as shown in [Figure 2](#).

This is an open access article under the terms of the [Creative Commons Attribution-NonCommercial License](#), which permits use, distribution and reproduction in any medium, provided the original work is properly cited and is not used for commercial purposes.

© 2023 The Authors. *Clinical Case Reports* published by John Wiley & Sons Ltd.

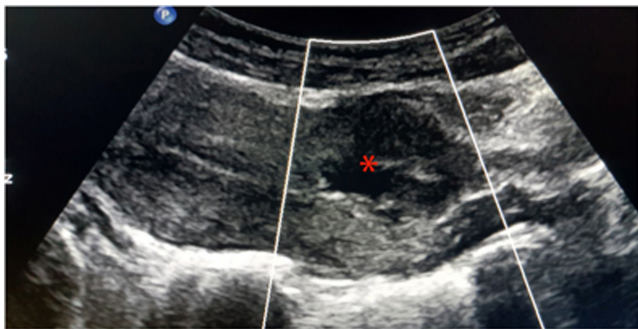


FIGURE 1 Trans vaginal ultrasonography shows an anechoic defect (red asterisk) in cesarean incision site.

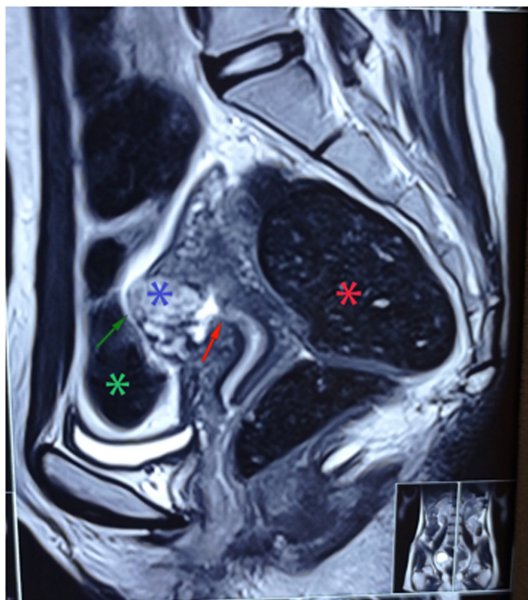


FIGURE 2 Axial image of magnetic resonance imaging (MRI) of abdomen/pelvis shows loss of myometrial continuity (red arrow) with blood clot (purple asterisk) and intact serosa (green arrow). Bowels (red asterisk) and bladder (green asterisk) are also seen.

She underwent laparotomy which revealed 2 × 2 cm bluish bulge at the previous scar site as shown in [Figure 3](#). Upon incising the bulge, brownish product of conception like material was revealed as depicted in [Figures 4 and 5](#). The scar tissue was removed as shown in [Figure 6](#) and the margins were freshened. The uterine cavity was difficult to visualize so, uterine sound was inserted and the uterine incision was closed in two layers as shown in [Figure 7](#). Histopathological examination of brownish material obtained after incising the bulge revealed to be the product of conception.

The postoperative period was uneventful and she was discharged on the 3rd postoperative day. During the follow-up examination, she is doing well 6 months after surgery.

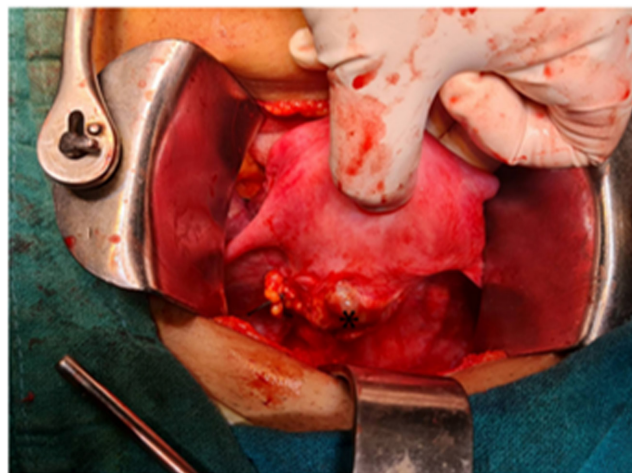


FIGURE 3 Per operative image showing bulge at the previous scar site (black asterisk).

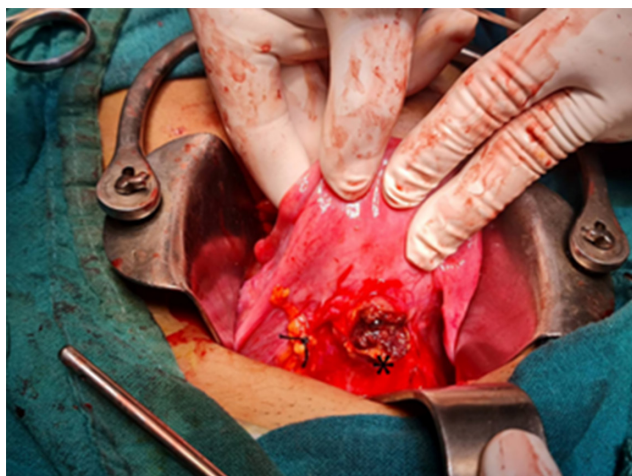


FIGURE 4 Per operative image showing brownish material exposed after incising the bulge present at the previous scar site (black asterisk).



FIGURE 5 Gross specimen of removed scar tissue.

3 | DISCUSSION

An isthmocele is described as a triangular hypoechoic zone at the site of the prior cesarean scar and constitutes an insufficient mending of the myometrium.⁷ The



FIGURE 6 Gross specimen of product of conception like mass removed after incising the bulge present at previous scar site.

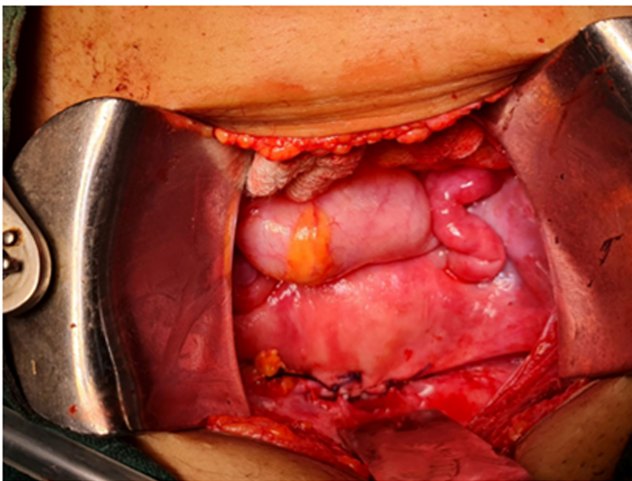


FIGURE 7 Per operative image showing uterus after the repair of isthmocele.

estimated incidence of pregnancy with a cesarean defect lies between 1/1800 and 1/2216, constituting 6.1% of all ectopic pregnancies with at least one prior cesarean section done.⁸ Although some women with an isthmocele are asymptomatic, others may have symptoms like postmenstrual spotting, contorted PV bleeding, ceaseless brown discharge, lengthened menstruation, chronic pelvic pain, and secondary infertility.^{9–12} In our case, the patient had irregular PV bleeding as a presenting symptom.

The invasion of blastocyst into the uterine musculature through the micro defect in hysterotomy scar has been hypothesized as the underlying pathophysiological mechanism for the development of pregnancy in an isthmocele.¹³ After analysis of 26 patients with cesarean scar defect, Gubbini et al.¹⁴ reported that the defects appeared anteriorly, near the right side. Our patient also had the defect anteriorly near the right side per trans vaginal ultrasound which was validated in laparotomy. This could be due to

the right inner edge of the myometrial incision being a blind spot because conventionally, the surgeon stands on the patient's right.¹⁴ Hence, to reduce interspaces when suturing the inner edge of the cesarean incision at uterine closure, more scrutiny should be given.¹⁴

Irregular PV bleeding and/or heavy menstrual bleeding associated with an isthmocele as present in our case can be due to reduced contractility in the lower uterine segment slowing the drainage of menstrual blood or the blood collecting in the cavity with unhurried subsequent drainage.^{15,16} Further various endometrial abnormalities such as division and breakdown of endometrium, protruding congested endometrium, and incorporation of endometrium within the scar can also lead to abnormal bleeding in cesarean scar defects.^{4,17} Slow seepage of menstrual blood as well as endometrial abnormalities can affect sperm swim-up and their survival and can lead to infertility together with interference to embryo implantation.¹¹

The most pivotal technical facet of an isthmocele is the correct discerning of the defect which usually requires a combination of surgery and one or more of various procedures such as transvaginal ultrasound, MRI, hysterosalpingography, hysteroscopy, and sonohysterography performed prior to surgery.^{7,8} Because of the rarity of the condition, there are no specific exhortations for therapeutic interventions.⁸ Treatment, whether medical or surgical, is based on taking consideration of the gestational age, the availability of therapeutic means, the patient's inclination for future fertility, the experience of the therapeutic team, and the complications of first-line therapy.⁸ Medical treatment focuses on the dispensation of methotrexate locally or systemically while surgical treatment options such as operative hysteroscopy, conventional or robotic-assisted laparoscopic surgery, laparotomy, and vaginal surgery have been described.^{8,18–21} Laparotomy and laparoscopy can permit complete excision of the scar and evacuation of trophoblastic tissue.⁸ Our patient underwent laparotomy with removal of scar tissue and product of conception, and finally closure of uterine incision.

There are studies which have highlighted the technique of closure of uterine incision as a risk factor for cesarean scar defect.^{22–25} Furthermore, multiple cesarean sections also help to increase the incidence of isthmus uteri.²⁶ The double layer uterine closure along with endometrial suturing can be considered as the finest way to avert scar defects.^{25,26}

There remains about a 5% chance of recurrence of cesarean scar pregnancy in women with a past history of cesarean scar ectopic pregnancy.²⁷ Time frame for initiating a new pregnancy in these women ranges from 2 months to 2 years.²⁷ Pregnancies have been described in women with a past history of cesarean scar ectopic pregnancy and since there is high chance of uterine rupture, elective cesarean

section is the preferred mode of delivery done as soon as acceptable pulmonary maturity is reached.²⁷

4 | CONCLUSION

With the rise in cesarean section rates throughout the world, there is likelihood of confronting unorthodox cesarean scar defects including rare entities like pregnancy occurring in the uterine niche. Early accurate diagnosis can be possible if isthmocele is kept in mind while dealing with post cesarean cases presenting with unexplained bleeding, thereby leading to appropriate diagnostic and therapeutic interventions.

AUTHOR CONTRIBUTIONS

Alina Tandukar (AT), Bishal Khaniya (BK), Suvana Maskey (SM), and Neebha Ojha (NO) = Study concept and provided counseling and surgical therapy to the patient. Alina Tandukar (AT) and Roshan Aryal (RA) = Collected all the required case information, images, slides, reports; reviewed the literature, and contributed in both writing and editing the manuscript. BK, SM, NO, and Dinesh Chataut (DC) = senior author and manuscript reviewer. All the authors read and approved the manuscript.

ACKNOWLEDGEMENTS

None.

FUNDING INFORMATION

Not available.

CONFLICT OF INTEREST

None to declare.

DATA AVAILABILITY STATEMENT

All the necessary data and materials are within the manuscript.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

ORCID

Roshan Aryal  <https://orcid.org/0000-0002-4689-9264>

REFERENCES

1. Bij de Vaate AJM, Brölmann HAM, van der Voet LF, van der Slikke JW, Veersema S, Huirne JAF. Ultrasound evaluation of the cesarean scar: relation between a niche and postmenstrual spotting. *Ultrasound Obstet Gynecol.* 2011;37(1):93-99.
2. Vikhareva Osser O, Valentin L. Clinical importance of appearance of cesarean hysterotomy scar at transvaginal ultrasonography in nonpregnant women. *Obstet Gynecol.* 2011;117(3):525-532.
3. Pędraszewski P, Wlazlak E, Panek W, Surkont G. Cesarean scar pregnancy – a new challenge for obstetricians. *J Ultrason.* 2018;18(72):56-62.
4. Morris H. Surgical pathology of the lower uterine segment caesarean section scar: is the scar a source of clinical symptoms? *Int J Gynecol Pathol.* 1995;14(1):16-20.
5. Futyma K, Gałczyński K, Romanek K, Filipczak A, Rechberger T. When and how should we treat cesarean scar defect – isthmocele? *Ginekolog pol.* 2016;87(9):664-668.
6. Antila-Långsjö RM, Mäenpää JU, Huhtala HS, Tomás EI, Staff SM. Cesarean scar defect: a prospective study on risk factors. *Am J Obstet Gynecol.* 2018;219(5):458.e1-e458.e8.
7. Monteagudo A, Carreno C, Timor-Tritsch IE. Saline infusion sonohysterography in nonpregnant women with previous cesarean delivery: the “niche” in the scar. *J Ultrasound Med.* 2001;20(10):1105-1115.
8. Rotas MA, Haberman S, Levгур M. Cesarean scar ectopic pregnancies: etiology, diagnosis, and management. *Obstet Gynecol.* 2006;107(6):1373-1381.
9. Borges LM, Scapinelli A, de Baptista DD, Lippi UG, Coelho Lopes RG. Findings in patients with postmenstrual spotting with prior cesarean section. *J Minim Invasive Gynecol.* 2010;17(3):361-364.
10. Wang CB, Chiu WWC, Lee CY, Sun YL, Lin YH, Tseng CJ. Cesarean scar defect: correlation between cesarean section number, defect size, clinical symptoms and uterine position. *Ultrasound Obstet Gynecol.* 2009;34(1):85-89.
11. Bij de Vaate AJM, van der Voet LF, Naji O, et al. Prevalence, potential risk factors for development and symptoms related to the presence of uterine niches following cesarean section: systematic review. *Ultrasound Obstet Gynecol.* 2014;43(4):372-382.
12. Uppal T, Lanzarone V, Mongelli M. Sonographically detected caesarean section scar defects and menstrual irregularity. *J Obstet Gynaecol.* 2011;31(5):413-416.
13. Maymon R, Halperin R, Mendlovic S, Schneider D, Herman A. Ectopic pregnancies in a caesarean scar: review of the medical approach to an iatrogenic complication. *Hum Reprod Update.* 2004;10(6):515-523.
14. 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;15(2):172-175.
15. Thurmond AS, Harvey WJ, Smith SA. Cesarean section scar as a cause of abnormal vaginal bleeding: diagnosis by sonohysterography. *J Ultrasound Med.* 1999;18(1):13-16; quiz 17-18.
16. Fabres C, Aviles G, De La Jara C, et al. The cesarean delivery scar pouch: clinical implications and diagnostic correlation between transvaginal sonography and hysteroscopy. *J Ultrasound Med.* 2003;22(7):695-700; quiz 701-702.
17. Drouin O, Bergeron T, Beaudry A, Demers S, Roberge S, Bujold E. Ultrasonographic evaluation of uterine scar niche before and after laparoscopic surgical repair: a case report. *AJP Rep.* 2014;4(2):e65-e68.

18. Xie H, Wu Y, Yu F, He M, Cao M, Yao S. A comparison of vaginal surgery and operative hysteroscopy for the treatment of cesarean-induced isthmocele: a retrospective review. *Gynecol Obstet Invest.* 2014;77(2):78-83.
19. Donnez O, Jadoul P, Squifflet J, Donnez J. Laparoscopic repair of wide and deep uterine scar dehiscence after cesarean section. *Fertil Steril.* 2008;89(4):974-980.
20. Florio P, Filippeschi M, Moncini I, Marra E, Franchini M, Gubbini G. Hysteroscopic treatment of the cesarean-induced isthmocele in restoring infertility. *Curr Opin Obstet Gynecol.* 2012;24(3):180-186.
21. Yalcinkaya TM, Akar ME, Kammire LD, Johnston-MacAnanny EB, Mertz HL. Robotic-assisted laparoscopic repair of symptomatic cesarean scar defect: a report of two cases. *J Reprod Med.* 2011;56(5-6):265-270.
22. Hamar BD, Saber SB, Cackovic M, et al. Ultrasound evaluation of the uterine scar after cesarean delivery: a randomized controlled trial of one- and two-layer closure. *Obstet Gynecol.* 2007;110(4):808-813.
23. Hayakawa H, Itakura A, Mitsui T, et al. Methods for myometrium closure and other factors impacting effects on cesarean section scars of the uterine segment detected by the ultrasonography. *Acta Obstet Gynecol Scand.* 2006;85(4):429-434.
24. Vikhareva Osser O, Valentin L. Risk factors for incomplete healing of the uterine incision after caesarean section. *BJOG.* 2010;117(9):1119-1126.
25. Yazicioglu F, Gökdoğan A, Kelekci S, Aygün M, Savan K. Incomplete healing of the uterine incision after caesarean section: is it preventable? *Eur J Obstet Gynecol Reprod Biol.* 2006;124(1):32-36.
26. Ofili-Yebovi D, Ben-Nagi J, Sawyer E, et al. Deficient lower-segment cesarean section scars: prevalence and risk factors. *Ultrasound Obstet Gynecol.* 2008;31(1):72-77.
27. Ben Nagi J, Helmy S, Ofili-Yebovi D, Yazbek J, Sawyer E, Jurkovic D. Reproductive outcomes of women with a previous history of cesarean scar ectopic pregnancies. *Hum Reprod.* 2007;22(7):2012-2015.

How to cite this article: Tandukar A, Aryal R, Khaniya B, Maskey S, Ojha N, Chataut D. Pregnancy in an isthmocele: A rare case from Nepal. *Clin Case Rep.* 2023;11:e06875. doi:[10.1002/ccr3.6875](https://doi.org/10.1002/ccr3.6875)