

Case Report,

Recurrent Isthmocele Following Second Cesarean Section Delivery: A Case Report and Management Strategies

Ghukasyan N.N.*

Maternity House of Erebouni Medical center Armenia.

*Corresponding author: Ghukasyan N.N.; norayrghukasyan2020@gmail.com

Received: 20 June 2024;

Revised: 1 July 2024;

Accepted: 14 July 2024;

Published: 23 July 2024

Abstract:

Background: Cesarean section (CS) rates are increasing globally, leading to heightened concern regarding associated adverse outcomes, including isthmocele formation. Isthmocele, a depression or defect in the anterior uterine wall at the CS scar site, can result in various complications, impacting women's quality of life and reproductive health. **Methods:** We present a case report of a 30-year-old woman who underwent emergency CS in 2019 and subsequently developed isthmocele. After experiencing lower abdominal pain, she underwent clinical and laboratory investigations followed by laparoscopic resection of the left ovary. Due to secondary infertility and the desire for future pregnancies, she underwent laparoscopic metroplasty. **Results:** Following metroplasty, the patient conceived naturally and had a successful pregnancy monitored as high-risk. She underwent cesarean section delivery without complications and was discharged postpartum. Subsequent ultrasound revealed the recurrence of isthmocele, highlighting the need for ongoing monitoring and intervention. **Conclusion:** This case underscores the importance of timely diagnosis and appropriate management of isthmocele, particularly in women desiring future pregnancies. Minimally invasive surgical techniques such as laparoscopic metroplasty can effectively treat isthmocele, facilitating successful pregnancies and improving maternal outcomes.

Keywords: caesarean scar defect, isthmocele, reproductive surgery, laparoscopy

Introduction

With the increasing rate of cesarean sections (CS) worldwide, attention to the adverse outcomes of this procedure is becoming more acute. Apart from immediate complications such as bleeding and infections, the rising frequency of CS is also associated with an increased risk of developing late complications, including isthmocele and placental accreta.

Isthmocele, one of the adverse consequences of CS, manifests as a depression or hypoechoic defect in the anterior uterine wall at the site of the CS scar. Although often asymptomatic, isthmocele can sometimes be accompanied by abnormal uterine bleeding and chronic pelvic pain. Factors contributing to its development include emergency surgical interventions, non-compliance with surgical techniques, low (cervical) hysterotomy, myometrial splitting, inadequate hemostasis leading to hematoma formation, as well as the use of low-quality suture materials and sparing "single-layer" closure of the uterine incision, pathological blood loss, hemostatic system disorders, repeated cesarean deliveries, and infectious-inflammatory complications (endometritis, endomyometritis) in the postpartum period.

Isthmocele outside of pregnancy is often underestimated, leaving patients untreated for extended periods despite significant deterioration in their quality of life due to the persistent manifestation of clinical symptoms such as suprapubic pain, profuse prolonged menstruation, postmenstrual bloody discharge, secondary infertility, and dyspareunia.

Infertility, placental adhesion or previa, scar divergence, uterine rupture, and ectopic pregnancy after CS can also be complications of this condition. Currently established risk factors for isthmocele

development include uterine retroflexion and multiple cesarean sections. However, factors such as low CS position, incomplete hysterotomy closure, early uterine wall adhesion, and genetic predisposition may also contribute to niche development. Since there are no definitive diagnostic criteria for isthmocele, several visualization methods can be used to assess uterine wall integrity and thus diagnose isthmocele. However, transvaginal ultrasound and sonohysterography with saline infusion have become specific, sensitive, and economically efficient methods for diagnosing isthmocele. Treatment includes clinical or surgical intervention depending on the defect size, presence of symptoms, secondary infertility, and plans for childbearing. Surgical treatment includes minimally invasive approaches using gentle methods such as hysteroscopic, laparoscopic, or transvaginal procedures, depending on the defect size.

Despite its potential impact on quality of life, isthmocele outside of pregnancy is often ignored, leaving patients untreated. Complications associated with isthmocele can range from infertility to uterine rupture, necessitating timely diagnosis and intervention. Visualization methods such as transvaginal ultrasound and sonohysterography are becoming increasingly important for diagnosing isthmocele.

Approaches to treatment vary depending on the size and symptoms of the defect, including conservative treatment and surgical interventions. Recent data suggest the effectiveness of hysteroscopic techniques and laparoscopic procedures in treating isthmocele. However, the choice of surgical interventions should be individualized, considering their implications for future reproductive function and pregnancy outcomes.

In this article, we present a case of isthmocele, describing its diagnosis, treatment, and management, including infertility issues and postpartum complications.

Case Description

We report a case of patient A.L., 30 years old, who had previously undergone emergency cesarean section due to fetal distress (2019). She presented to Erebuni Medical Center on March 18, 2022, complaining of lower abdominal pain, predominantly on the left side, for the past 2 days.

Preliminary diagnosis: acute abdominal pain and partial torsion of the left ovary.

After conducting all clinical and laboratory investigations, surgical treatment was proposed to the patient. It is worth noting that during the ultrasound examination with the participation of a gynecologist, a pronounced isthmocele was detected - a scar defect of the uterus measuring 2.0x1.3 cm, and cystic lesions of anechoic structure measuring 0.6x0.6 cm were identified on the anterior wall of the myometrium (which was not calm) (**Figure. 1**).

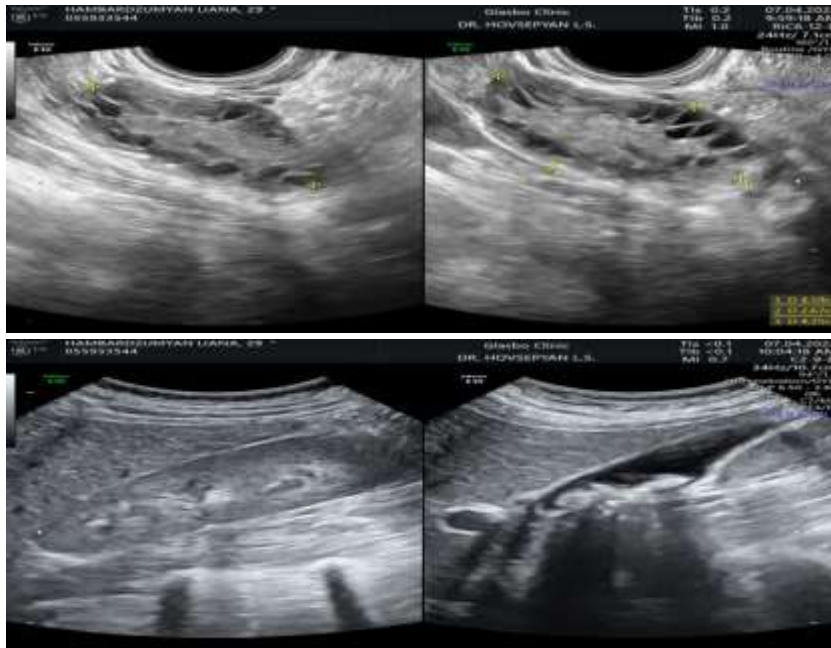


Figure 1 - Uterine wall defect resembling a sail before metroplasty

The patient was counseled regarding the risks associated with this condition, available treatment options, and their complications. Written informed consent for reporting the case was obtained from the patient prior to the procedure. The patient underwent laparoscopic resection of the left ovary with abdominal cavity sanitation and drainage.

Due to the patient's desire to have more children and the presence of secondary infertility caused by isthmocele (Figure. 2), the

obstetrician-gynecologist referred her to a reproductive specialist who began monitoring and managing her case. After a thorough examination and consultation by the reproductive specialist, it was proposed, in order to prevent reproductive losses (miscarriage, abnormal placentation, uterine rupture in the third trimester), to restore uterine integrity through metroplasty.

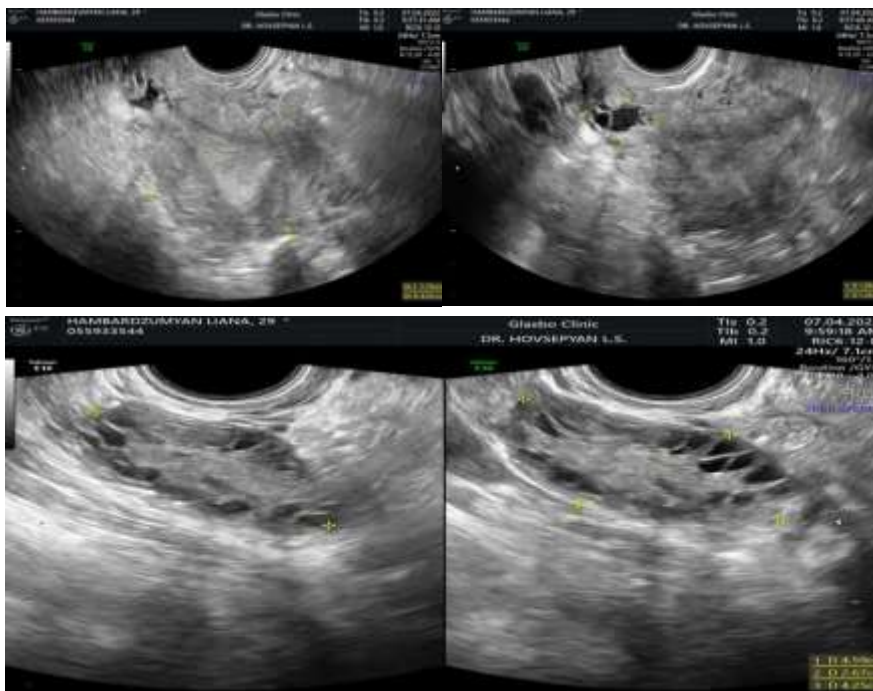




Figure 2. Image of isthmocele.

After the postoperative recovery period, due to infertility issues, the patient was offered laparoscopic hysteroscopy and metroplasty. As known, in such cases, hormonal therapy is advisable to consider as symptomatic treatment in women who are not planning conception and have no contraindications. In cases of ineffectiveness or contraindications to medical treatment, the possibility of surgical intervention is considered depending on the

severity of symptoms, including infertility, desire to preserve the uterus, defect size, and residual myometrial thickness measurement. Hysteroscopy was performed, assessing myometrial inadequacy, isthmocele laparoscopically, and metroplasty was conducted. The course was uneventful, and the expected response was obtained (Figure. 3).



Figure 3 - Pregnancy and myometrial condition at different stages.

After undergoing hysteroscopy and metroplasty on April 3, 2022, the patient was under the observation of a reproductive specialist for 6 months and then placed under the care of an obstetrician-gynecologist 3 months later with a normally developing pregnancy at 10 weeks. The pregnancy was monitored as a high-risk

pregnancy at Erebuni Medical Center. Throughout the pregnancy and postpartum period, she was administered vaginal progesterone until 34 weeks. The focus was primarily on the condition of the sutures, and apart from the protocol-driven 3-4D scanning, there were mostly no issues, even without hospitalization.

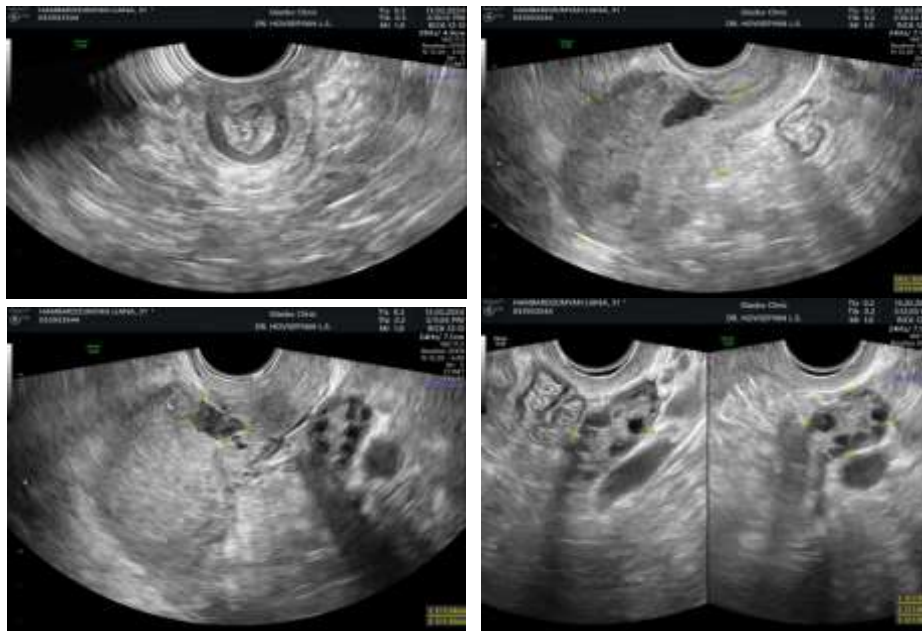


Figure 4 - Myometrium after childbirth

At 37 weeks of pregnancy, the patient started experiencing regular contractions, leading to cesarean section delivery on February 1, 2024. A healthy baby boy was born, weighing 3100g, measuring 50cm, with an Apgar score of 8.8.

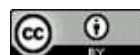
The postoperative period was uneventful, with no bleeding and normal uterine contractions. She was discharged with the baby. On a follow-up ultrasound examination 42 days later, an isthmocele

was observed (Fig. 4). Currently, there are no clinical symptoms, and further management will depend on the patient's quality of life and her preferences. Nothing is impossible in obstetrics and gynecology; with the right protocols and interventions, we can effectively improve the integrity of the myometrium, thereby restoring fertility and enabling women to conceive again.

References

- [1] Kulshrestha V, Agarwal N, Kachhawa G. Post-caesarean Niche (Isthmocele) in Uterine Scar: An Update. *J Obstet Gynaecol India*. 2020; 70:6:440-446. <https://doi.org/10.1007/s13224-020-01370-0>
- [2] Gulz M, Imboden S, Nirgianakis K, Siegenthaler F, Rau TT, Mueller MD. Endometriosis and isthmocele: common or rare? *J Clin Med*. 2022; 11:5:1158. <https://doi.org/10.3390/jcm11051158>
- [3] Al Naimi A, Wolnicki B, Mouzakiti N, Reinbach T, Louwen F, Bahlmann F. Anatomy of the sonographic post-caesarean uterus. *Arch Gynecol Obstet*. 2021; 304:6:1485-1491. <https://doi.org/10.1007/s00404-021-06074-y>
- [4] Vervoort AJ, Uittenbogaard LB, Hehenkamp WJ, Brölmann HA, Mol BW, Huirne JA. Why do niches develop in caesarean uterine scars? Hypotheses on the aetiology of niche development. *Hum Reprod*. 2015; 30:12:2695-2702. <https://doi.org/10.1093/humrep/dev240>
- [5] Kremer TG, Ghiorzi IB, Dibi RP. Isthmocele: an overview of diagnosis and treatment. *Rev Assoc Med Bras (1992)*. 2019; 65:5:714-721. <https://doi.org/10.1590/1806-9282.65.5.714>
- [6] Bamberg C, Hinkson L, Dudenhausen JW, Bujak V, Kalache KD, Henrich W. Longitudinal transvaginal ultrasound evaluation of cesarean scar niche incidence and depth in the first two years after single- or double-layer uterotomy closure: a randomized controlled trial. *Acta Obstet Gynecol Scand*. 2017;96:12:1484-1489. <https://doi.org/10.1111/aogs.13213>
- [7] Budny-Winska J, Zimmer-Stelmach A, Pomorski M. Two- and three-dimensional transvaginal ultrasound in assessment of the impact of selected obstetric risk factors on cesarean scar niche formation: the case-controlled study. *Ginekol Pol*. 2021;92:5:378-382. <https://doi.org/10.5603/GP.a2021.0024>
- [8] Park IY, Kim MR, Lee HN, Gen Y, Kim MJ. Risk factors for Korean women to develop an isthmocele after a cesarean section. *BMC Pregnancy Childbirth*. 2018;18:1:162. <https://doi.org/10.1186/s12884-018-1821-2>
- [9] Antila-Långsjö R, Mäenpää JU, Huhtala H, Tomás E, Staff S. Comparison of transvaginal ultrasound and saline contrast sonohysterography in evaluation of cesarean scar defect: a prospective cohort study. *Acta Obstet Gynecol Scand*. 2018;97:9:1130-1136. <https://doi.org/10.1111/aogs.13367>
- [10] Yılmaz Baran Ş, Kalaycı H, Doğan Durdağ G, Yetkinel S, Alemdaroğlu S, Çok T, Bulgan Kılıçdağ E. Single- or double-layer uterine closure techniques following cesarean: A randomized trial. *Acta Obstet Gynecol Scand*. 2021;100:3:531-537. <https://doi.org/10.1111/aogs.14018>
- [11] Bij de Vaate AJ, van der Voet LF, Naji O, Witmer M, Veersema S, Brölmann HA, Bourne T, Huirne JA. 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. <https://doi.org/10.1002/uog.13199>
- [12] Gonser M. Re: Prevalence, potential risk factors for development and symptoms related to the presence of uterine niches following cesarean section: systematic review. *Ultrasound Obstet Gynecol*. 2014;44:3:371. <https://doi.org/10.1002/uog.14631>

- [13] Stegwee SI, van der Voet LF, Ben AJ, de Leeuw RA, van de Ven PM, Duijnhoven RG, Bongers MY, Lambalk CB, de Groot C, Huirne J; 2Close study group. Effect of single-versus double-layer uterine closure during caesarean section on postmenstrual spotting (2Close): multicentre, double-blind, randomized controlled superiority trial. *BJOG*. 2021;128:5:866-878. <https://doi.org/10.1111/1471-0528.16472>
- [14] Kataoka S, Tanuma F, Iwaki Y, Iwaki K, Fujii T, Fujimoto T. Comparison of the primary cesarean hysterotomy scars after single- and double-layer interrupted closure. *Acta Obstet Gynecol Scand*. 2016;95:12:1352-1358. <https://doi.org/10.1111/aogs.13015>
- [15] Di Spiezio Sardo A, Saccone G, McCurdy R, Bujold E, Bifulco G, Berghella V. Risk of cesarean scar defect following single- vs double-layer uterine closure: systematic review and meta-analysis of randomized controlled trials. *Ultrasound Obstet Gynecol*. 2017;50:5:578-583. <https://doi.org/10.1002/uog.17401>
- [16] ErKayiran U, Arslanca T. Comparative analysis of classical primary continuous and novel technique uterine suturing methods on uterine scar formation after caesarean section: a prospective clinical study. *Ginekol Pol*. 2022;93:7:552-557. <https://doi.org/10.5603/GP.a2022.0022>
- [17] Pomorski M, Fuchs T, Rosner-Tenerowicz A, Zimmer M. Morphology of the cesarean section scar in the non-pregnant uterus after one elective cesarean section. *Ginekol Pol*. 2017;88:4:174-179. <https://doi.org/10.5603/GP.a2017.0034>
- [18] Marchand GJ, Masoud A, King A, Ruther S, Brazil G, Ulibarri H, Parise J, Arroyo A, Coriell C, Goetz S, Christensen A, Sainz K. Effect of single- and double-layer cesarean section closure on residual myometrial thickness and isthmocele — a systematic review and meta-analysis. *Turk J Obstet Gynecol*. 2021;18:4:322-332. <https://doi.org/10.4274/tjod.galenos.2021.71173>
- [19] Sholapurkar SL. Etiology of cesarean uterine scar defect (Niche): detailed critical analysis of hypotheses and prevention strategies and peritoneal closure debate. *J Clin Med Res*. 2018;10:3:166-173. <https://doi.org/10.14740/jocmr3271w>
- [20] Dawood AS, Elgergawy A, Elhalwagy A, Ataallah WM, Elbohoty SB, Elshwaikh SL, Elsokary AA, Elkhyat AM, Elbadry AT, Abbas AM. The impact of mechanical cervical dilatation during elective cesarean section on postpartum scar integrity: a randomized double-blind clinical trial. *Int J Womens Health*. 2019;11:23-29. <https://doi.org/10.2147/IJWH.S188628>
- [21] Kamel R, Eissa T, Sharaf M, Negm S, Thilaganathan B. Position and integrity of uterine scar are determined by degree of cervical dilatation at time of cesarean section. *Ultrasound Obstet Gynecol*. 2021;57:3:466-470. <https://doi.org/10.1002/uog.22053>
- [22] Roberge S, Demers S, Girard M, Vikhareva O, Markey S, Chaillet N, Moore L, Paris G, Bujold E. Impact of uterine closure on residual myometrial thickness after cesarean: a randomized controlled trial. *Am J Obstet Gynecol*. 2016;214:4:507.e1-507.e6. <https://doi.org/10.1016/j.ajog.2015.10.916>
- [23] Bennich G, Rudnicki M, Wilken-Jensen C, Lousen T, Lassen PD, Wøjdemann K. Impact of adding a second layer to a single unlocked closure of a cesarean uterine incision: randomized controlled trial. *Ultrasound Obstet Gynecol*. 2016;47:4:417-422. <https://doi.org/10.1002/uog.15792>
- [24] Karakuş R, Karakuş SS, Güler B, Ünver G, Özkaya E. Myometrial thickness overlying cesarean scar pregnancy is significantly associated with isthmocele formation in the third month of the postoperative period. *Turk J Obstet Gynecol*. 2021;18:1:37-43. <https://doi.org/10.4274/tjod.galenos.2021.65288>



Open Access This article is licensed under a Creative Commons Attribution 4.0 International License, which permits use, sharing, adaptation, distribution and reproduction in any medium or format, as long as you give appropriate credit to the original author(s) and the source, provide a link to the Creative Commons license, and indicate if changes were made. The images or other third party material in this article are included in the article's Creative Commons license, unless indicated otherwise in a credit line to the material. If material is not included in the article's Creative Commons license and your intended use is not permitted by statutory regulation or exceeds the permitted use, you will need to obtain permission directly from the copyright holder. To view a copy of this license, visit <http://creativecommons.org/licenses/by/4.0/>.

© The Author(s) 2024