

## Pouch of sorrow: Undiagnosed isthmocele – presenting as secondary infertility of 11 years in a middle age female

Sayema<sup>1</sup>, Aiman Ibrahimi<sup>2</sup>, Divyashree Koppal<sup>2</sup>, Shagufta Wahab<sup>3</sup>

From <sup>1</sup>Assistant Professor, <sup>2</sup>23<sup>rd</sup> Year Junior Resident, <sup>3</sup>Professor, Department of Radiodiagnosis, Jawaharlal Nehru Medical College and Hospital, Aligarh Muslim University, Aligarh, Uttar Pradesh, India

### ABSTRACT

In the past decade, the rate of cesarean sections has doubled leading to the more frequent occurrence of cesarean-related complications. Cesarean scar site diverticulum or isthmocele is one such complication that is defined as an outpouching of the anterior uterine wall at the cesarean scar site in continuity with the endometrial cavity, creating a wedge-shaped defect of variable depth. We report a case of a middle-aged female with secondary infertility for the last 11 years with one live issue 12 years back through a cesarean section at term. She had undergone an array of investigations for infertility for the past 5 years. She was diagnosed as having an isthmocele and managed with surgical repair of the defect at our hospital with subsequent assisted conception 8-month post-surgical repair. The importance of diagnosing a cesarean scar diverticulum cannot be understated not only because it is a surgically treatable cause of infertility but also because if such patients conceive, they may land in uterine rupture, placenta accreta, or scar ectopic pregnancy which can complicate into life-threatening situations.

**Key words:** Cesarean scar diverticulum, Isthmocele, Post-cesarean scar defect, Secondary infertility

In the past decade, the rate of cesarean sections has doubled leading to the more frequent occurrence of cesarean-related complications [1]. Even though it is considered a safe procedure, many acute and chronic complications are frequently seen such as endometritis, wound infection, wound disruption, thrombophlebitis, and uterine scar dehiscence in a subsequent pregnancy [2]. Cesarean scar diverticulum is one such complication. It can be asymptomatic in many patients [3]. Isthmocele can act as a reservoir for the menstrual blood in which the menstrual fluid accumulates within the diverticulum and later presents as intermenstrual and postmenstrual bleeding. Other common clinical presentations are dysmenorrhea, dyspareunia, chronic pelvic pain, and even secondary infertility due to altered internal milieu of the endometrial cavity and scarring at the site.

### CASE REPORT


A 33-year-old female presented to the OPD of obstetrics and gynecology with complaints of secondary infertility for the past 11 years. Her menstrual history revealed irregular menstruation with postmenstrual spotting off and on. Her obstetric history

revealed one live issue with term delivery through cesarean section 12 years back in a private hospital for obstructed labor. The patient also had complaints of dysmenorrhea.

She was vitally stable with general and systemic examination findings within normal limits. Per vaginal and per speculum examination were normal with normal uterus and adnexa.

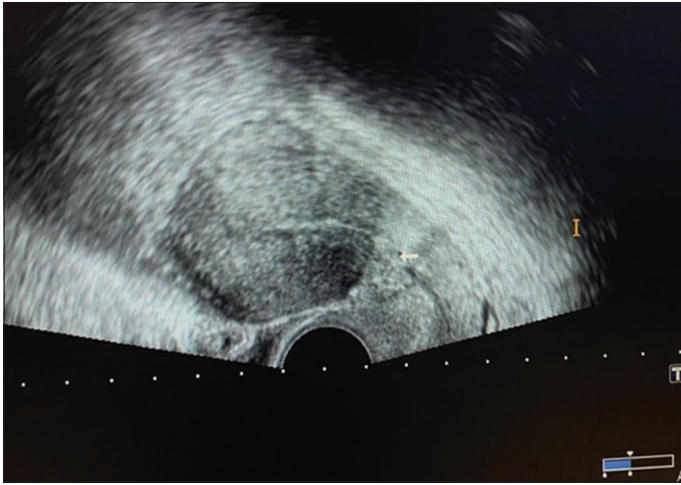
Her routine blood investigations (complete blood count, urine analysis, liver and renal function tests, and thyroid profile) revealed no abnormalities. She was carrying a previous hysterosalpingogram study report (6 months old) and a transabdominal ultrasound (USG) whole abdomen report, both revealing no significant diagnostic abnormality.

The patient came to the USG section of the Department of Radiodiagnosis where a transvaginal USG scan was performed. She was in day 5 of her cycle as per her last menstrual period. It revealed thinning of the overlying anterior uterine wall in the lower segment at the cesarean scar site up to 2 mm (Fig. 1). Due to the clinical history of intermenstrual bleeding, the possibility of isthmocele at the cesarean scar site was raised. The patient subsequently went for a pelvic magnetic resonance imaging (MRI) 3 days later in our department. MRI revealed thinning of myometrium at the cesarean scar site with the altered signal minimal fluid collection, bulging as a diverticulum anteriorly

Access this article online	
Received - 24 November 2022 Initial Review - 06 December 2022 Accepted - 09 January 2023	Quick Response code 
DOI: 10.32677/ijcr.v9i1.3734	

**Correspondence to:** Dr. Sayema, Department of Radiodiagnosis, Jawaharlal Nehru Medical College and Hospital, Aligarh Muslim University, Aligarh, Uttar Pradesh, India. E-mail- sayema6@gmail.com

© 2023 Creative Commons Attribution-NonCommercial 4.0 International License (CC BY-NC-ND 4.0).



**Figure 1:** (a) Transvaginal ultrasound image of uterus in mid sagittal view where white arrow points to the cesarean scar site diverticulum (empty at time of scanning) arising and directly in continuation with endometrial cavity

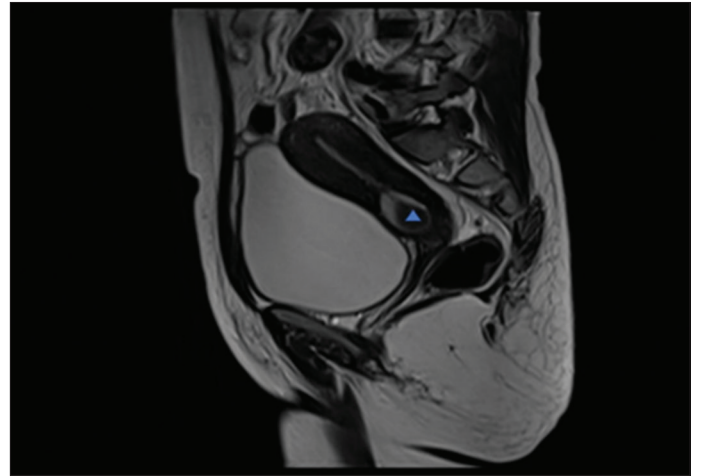
from the anterior wall of the lower uterine segment at the cesarean scar. The collection in the outpouching was T1 hyperintense and T2/PD hypointense with blooming artifacts on GRE sequences suggestive of altered blood products (Figs. 2 and 3). There was a 2mm thickness of intact myometrium covering the outpouching anteriorly. The diagnosis of cesarean scar site diverticulum with thinning of scar up to 2 mm and minimal altered fluid collection (hemorrhagic contents) was given.

The patient was taken up for the elective abdominal laparoscopic surgical repair of the diverticulum. Pre-operative findings revealed a small outpouching at the cesarean scar site at the uterine isthmus with residual thinned-out myometrium. On transverse incision over the diverticulum, it revealed brownish-black altered fluid (~10 ml) which was aspirated. The diverticulum was dissected and the defect was removed and repaired with 2–0 absorbable sutures.

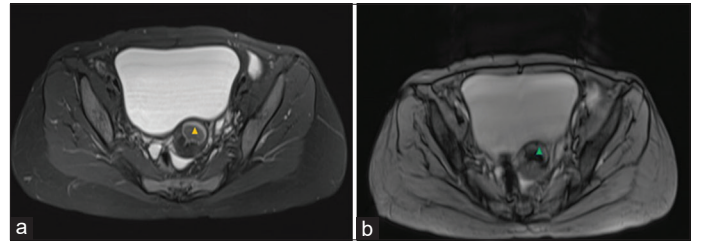
The post-operative period was uneventful. There was a significant improvement in the patient's symptoms. Intermenstrual bleeding completely resolved in the next menstrual cycle. No collection was seen on follow-up transvaginal sonography (TVS) 1 month later. Furthermore, 8-month post-operative, the patient successfully conceived through the assisted reproductive method (ovulation induction agents followed by intrauterine insemination).

## DISCUSSION

Due to the increasing number of cesarean sections and better diagnostic modalities available, the detection of complications related to cesarean section are on a rising trend in the last decade. The commonly associated complications include endometritis, wound infection, wound disruption, and thrombophlebitis. Common chronic or late complications include retained products of conception, adhesions, abdominal wall endometriosis, scar site ectopic pregnancy, cesarean scar diverticulum/defect, and placenta accreta [4]. However, the two most common late



**Figure 2:** Parasagittal MRI pelvis T2 Wt. image (blue triangle denotes altered signal intensity T2 hypointense collection lined by endometrial lining bulging at the cesarean scar site with thinning of overlying myometrium anteriorly)



**Figure 3:** (a) Axial MRI pelvis PD image at the level of lower uterine segment (yellow triangle denotes cesarean scar site diverticulum with thin but intact myometrium overlying); (b) Axial MRI pelvis GRE image at the level of lower uterine segment (green triangle denotes cesarean scar site diverticulum with contents of the diverticulum showing blooming/susceptibility artifact suggesting altered blood products)

complications of cesarean section are adhesions and previous cesarean scar defects.

The scar defect was present in 24–69% of women evaluated with TVS. It can lead to other serious complications such as uterine rupture while performing dilatation and curettage, insertion of intrauterine contraceptive devices, and myomectomy. There are many risk factors leading to the development of post-cesarean scar defects. These might be patient-related or related to the procedure. The patient-related risk factors include a retroverted uterus and the increasing age of the patient. Increased duration of active labor (more than 5 h) and cervical dilatation of more than 5 cm at the time of delivery are labour-related non-modifiable procedural risk factors. Modifiable procedural risk factors include surgical techniques such as low incision near the internal os, non-inclusion of the endometrium during uterine repair, single-layer closure, and inadequate healing of the cesarean incision [3].

The post-cesarean scar defect or isthmocele has been defined as the myometrial indentation of at least 2 mm at the scar site [5]. There has been a report suggesting approximately 50% of women who underwent cesarean section had cesarean scar niche on TVS, sonohysterography, and hysterothography [6]. The severity of complications depends on various factors such as

the size of the niche. When a niche is having a depth of at least 50–80% of the anterior myometrium or underlying myometrial thickness  $<2.2$  mm on TVS and  $\leq 2.5$  mm on sonohysterography is considered a large niche [7]. Most patients with cesarean scar defects are asymptomatic. However, it can cause chronic pelvic/abdominal pain, menometrorrhagia, and dyspareunia. The proportion of symptoms is directly related to the size of the defect. The larger the defect, the greater the clinical complaints by the patient [8]. In pregnant patients, the scar can expand and may rupture. In a few patients, if implantation occurs at the scar site, abnormal placentation, and rupture may occur [9].

The cesarean scar defect can rarely lead to secondary infertility. The reason for this is not clearly understood. This might be due to the collection of menstrual fluid in the sac which alters the internal milieu of the endometrial canal and can act as a site of inflammation and subsequent scarring [10].

TVS is the first imaging modality performed in almost all gynecological conditions. The diagnosis is inferred by the demonstration of a niche which is an anechoic area at the site of the cesarean scar with variable shape and size of the sac. Few studies have suggested that the early follicular phase is the best for sonographic evaluation as the lining of the endometrium is thin and therefore, the niche could be easily identified [11]. The best time to identify the pouch with sonography is during the bleeding episode, usually a few days after the menses, because the principle symptom is postmenstrual or intermenstrual spotting [12]. The shape of an isthmocele can be easily demonstrated by TVS by the instillation of saline or gel. The instillation of gel is better than saline as it provides a more stable acoustic window and being more viscous it is less leaky. The niche can be classified by its shape into triangular, semi-circular, rectangular, circle, droplet, and inclusion cyst [11].

MRI has advantages over transvaginal ultrasound as it gives a really greater contrast for the soft tissue of the pelvis and reduces interobserver variability in image acquisition. On MRI, an isthmocele may appear as an abnormal outline of the anterior uterine wall at the cesarean scar site which can be seen on both external and internal sides of the scar site and appears as semi-circular or triangular defect and hyperintense on sagittal T2-weighted images. The appearance may vary depending on the contents that are lodged within the niche. MRI also helps in differentiating uterine rupture from uterine dehiscence by delineating the intact overlying serosa of the uterus over it [13,14].

## CONCLUSION

Seeing the rising trend of deliveries by cesarean section, the complications related to it are also on the rise. This case report describes the clinical and imaging findings of one such complication of cesarean delivery which can lead to serious consequences including secondary infertility. TVS is the primary imaging modality that demonstrate an anechoic area at the site of the cesarean scar with thinned-out myometrium. MRI helps

to consolidate the diagnosis by demonstrating a T2 hyperintense semi-circular or triangular defect at the cesarean scar site and also altered signal intensity of the contents of this diverticulum which are often hemorrhagic.

## AUTHOR'S CONTRIBUTION

First and second authors were involved in patient diagnosis, management, and case report writing, first and third authors involved in conception of the article writing, editing, and final approval of manuscript.

## REFERENCES

1. Bhatia M, Banerjee K, Dixit P, Dwivedi LK. Assessment of variation in cesarean delivery rates between public and health facilities in India from 2005 to 2016. *JAMA Netw Open* 2020;3:e2015022.
2. Patrick D. A simple checklist for preventing major complications associated with cesarean delivery. *Obstet Gynecol* 2010;116:1393-6.
3. Rosa F, Perugini G, Schettini D, Romano N, Romeo S, Podestà R, *et al.* Imaging findings of cesarean delivery complications: Cesarean scar disease and much more. *Insights Imaging* 2019;10:98.
4. Rodgers SK, Kirby CL, Smith RJ, Horrow MM. Imaging after cesarean delivery: Acute and chronic complications. *Radiographics* 2012;32:1693-712.
5. 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;10:166-73.
6. Roberge S, Boutin A, Chaillet N, Moore L, Jastrow N, Demers S. Systematic review of cesarean scar assessment in the nonpregnant state: Imaging techniques and uterine scar defect. *Am J Perinatol* 2012;29:465-71.
7. Vervoort AJ, Uittenbogaard LB, Hehenkamp WJ, Brolmann HA, Mol BW, Huime JA. Why do niches develop in cesarean uterine scars? Hypotheses on the aetiology of niche development. *Hum Reprod* 2015;30:2695-702.
8. Uppal T, Lanzarone V, Mongelli M. Sonographically detected cesarean section scar defects and menstrual irregularity. *J Obstet Gynaecol* 2011;31:413-6.
9. Szkodziak P, Stepniak A, Czuczwar P, Szkodziak F, Paszkowski T, Woźniak S. Is it necessary to correct a cesarean scar defect before a subsequent pregnancy? A report of three cases. *J Int Med Res* 2019;47:2248-55.
10. Tanimura S, Funamoto H, Hosono T, Shitano Y, Nakashima M, Ametani Y, *et al.* New diagnostic criteria and operative strategy for cesarean scar syndrome: Endoscopic repair for secondary infertility caused by cesarean scar defect. *J Obstet Gynaecol Res* 2015;41:1363-9.
11. De Vaate AJ, Brölmann HA, Van der voet LF, Van der Slikke JW, Veersema S, Huime JA. Ultrasound evaluation of the cesarean scar: Relation between a niche and postmenstrual spotting. *Ultrasound Obstet Gynecol* 2011;37:93-9.
12. Maldjean C, Milestone B, Schnall M, Smith R. MR appearance of uterine dehiscence in post-cesarean section patient. *J Comput Assist Tomogr* 1998;22:738-41.
13. Chen Y, Chang Y, Yao S. Transvaginal management of cesarean scar section diverticulum: A novel surgical treatment. *Med Sci Monit* 2014;20:1395-9.
14. Van der Voet LF, Vervoort AJ, Veersema S, BijdeVaate AJ, Brölmann HA, Huime JA. Minimally invasive therapy for gynaecological symptoms related to a niche in the caesarean scar: A systematic review. *BJOG* 2014;121:145-56.

*Funding: Nil; Conflicts of interest: Nil.*

**How to cite this article:** Sayema, Ibrahim A, Koppal D, Wahab S. Pouch of sorrow: Undiagnosed isthmocele – presenting as secondary infertility of 11 years in a middle age female. *Indian J Case Reports*. 2023;9(1):14-16.