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Case Report

Chronic abscess in isthmocele: a rare entity

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ABSTRACT

Uterine isthmocele, also known as caesarean scar defect or uterine niche is a triangular defect in the anterior uterine wall at the site of the previous caesarean scar, with its base communicating with the uterine cavity. It represents an inadequate healing of the myometrium following caesarean section. Transvaginal ultrasound (TVS), saline infusion sonohysterogram (SIS), hysterosalpingogram, hysteroscopy, and MRI are various modalities to make a confirmatory diagnosis. Medical or surgical management is undertaken depending on the size and type of defect. The aim is to manage symptomatic patients. But chronic presentation of isthmocele is very rare and hence difficult to diagnose. Hereby presenting this case report, where the isthmocele was presented as a chronic abscess. Hence reporting this case for early suspicion of isthmocele and prompt management.

Keywords: Caesarean section, Myometrial defect, Isthmocele, Chronic abscess

INTRODUCTION

Uterine isthmocele is one of the late complications of caesarean deliveries. For the first time, Hugh Morris described "Isthmocele" in 1995 as a defect on the anterior wall of the uterine isthmus located at the site of previous caesarean scar. It was also referred to as "caesarean scar defect" or "niche." [1] It can present as Chronic pelvic pain, Heavy menstrual bleeding, post-menstrual spotting or secondary infertility and associated with a series of gynaecological and obstetrical problems.[2] In the last decade, as the rates of caesarean sections are constantly increasing worldwide, it is important to be aware of this entity so as to make an early diagnosis.

This would enable the clinicians to timely diagnose and manage effectively. It is extremely rare for isthmocele to remain quiescent for many years and to present in peri-menopausal age as persistent discharge PV. We report one such case of Uterine isthmocele in a peri-menopausal woman who presented with HMB, postmenstrual discharge PV and chronic pelvic pain.

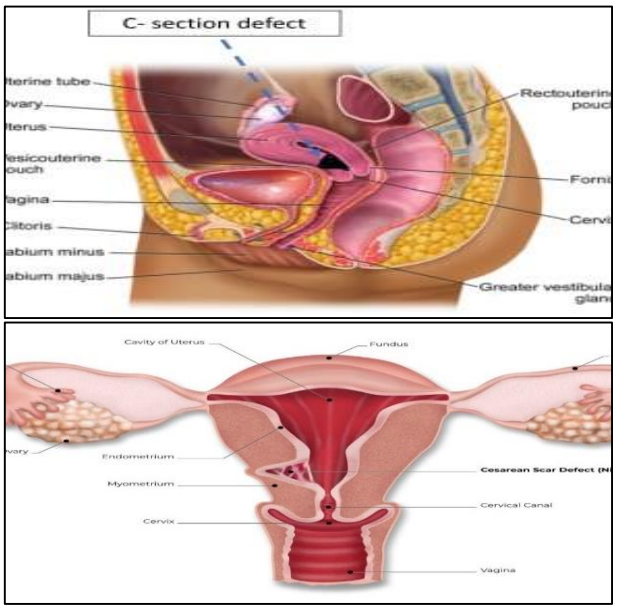

Figure 1: Sagittal and coronal view of isthmocele (C-section scar defect).

CASE REPORT

A 51 year old multiparous women with previous 2 caesarean sections, presented with complaints of chronic pelvic pain, HMB, prolonged intermenstrual spotting and persistent discharge PV of 1 year duration. She was treated at local hospital but as symptoms were nonresponsive to medical management, she was referred to the tertiary care medical college, Thiruvananthapuram, Kerala, with a provisional diagnosis of chronic pelvic inflammatory disease. She has H/o HMB lasting 2-3 weeks with usage of 8-9 pads per day, associated with dysmenorrhea since 1 year. She also has persistent vaginal discharge not responding to antibiotics and vaginal tablets. She has h/o 2 caesarean sections (27 and 24 years back), first caesarean was done for failed trial with h/o prolonged labour. Second CS done in view of prev caesarean. Since her second CS, she has h/o dyspareunia, congestive dysmenorrhea and her cycles were irregular with on and off intermenstrual bleeds for which she was symptomatically treated from local hospitals.

Over years she also developed persistent discharge PV. Since her symptoms did not improve, she was referred to medical college, trivandrum with an USG report showing-

USS (20/3/23)- Ut 5×5×11 cm with endometrial cavity distended and distorted by a heterogenous collection of 4×2.7 cm either hematometra or pyometra.

On clinical examination at our centre, patient was afebrile but pallor ++(Hb 7 gm%) was present, other vitals were stable. Per abdominal examination revealed a Pfannenstiel scar of prev 2 CS, but no mass or tenderness on palpation. On speculum examination a healthy cervix with slight blood-stained mucoid discharge was present. On bimanual pelvic examination, uterus was bulky and a cystic tender mass of 5×6 cm felt anterior and to the right side of body.

MRI was taken to define the pelvic pathology, MRI report revealed 'A thick walled cavitated lesion measuring 5.6×4.4×5.8 cm abutting the right lateral uterine wall at the junction of uterine body and cervix and its cavity communicating with lower endometrial cavity via a defect in right lateral wall of it. The cavity shows heterogenous $T2W_1$ hyperintense, and $T1W_1$ hypointense fluid collection showing significant diffusion restriction within.

The cavity shows thick walls with thickness of 17.4 mm superiorly. No diffusion restriction/ discernible focal lesion seen in cavity wall/uterine body / cervix. No collection within the endometrial cavity. Uterus measured 5.1×5.7×11.5 cm with endometrial thickness 5.6 mm.

No myometrial lesion, junctional zone appears normal, no focal cervical or vaginal lesions, B/L ovaries appear normal in size and signal. With these features a provisional diagnosis of chronic thick wall abscess communicating with lower endometrial cavity was made.

Possibilities of-chronic isthmocele, sub-serous or broad ligament fibroid with cystic degeneration communicating with lower uterine cavity and accessory and cavitated uterine mass with lower endometrial communication.

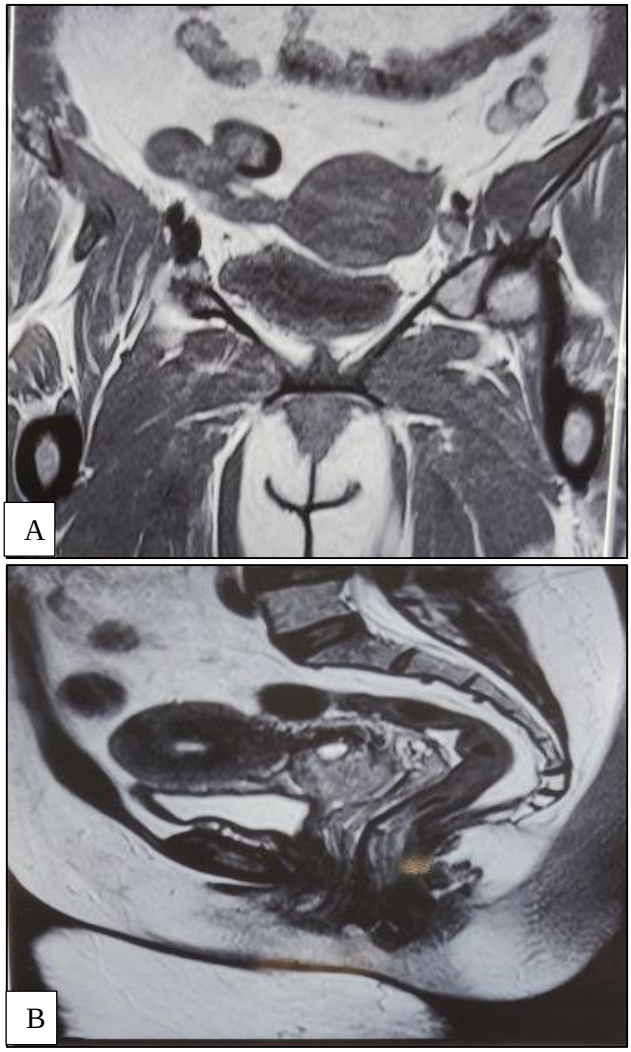

Figure 2: MRI showing thick wall abscess anterior to lower uterine segment and communicating to endometrial cavity (Coronal view and sagittal view).

Decision for laparotomy was taken after correction of anemia and a course of broad spectrum antibiotics. Intra operative findings were body of uterus, tubes and ovary normal.

A 5×4 cm was seen anterior and below the uterovesical fold, UV fold dissected, bladder densely adherent to the underlying bulge, sharp dissection was done to separate the adhesions. Meanwhile the cystic ruptured and thick pus drained. The mass was now clearly delineated, the base of which was attached to right side of isthmus. The sac was dissected from surrounding adhesion and proceeded with total hysterectomy with B/L salpingo-oophorectomy with the base of the sac still attached to the uterus. Features was typical of uterine isthomocele.

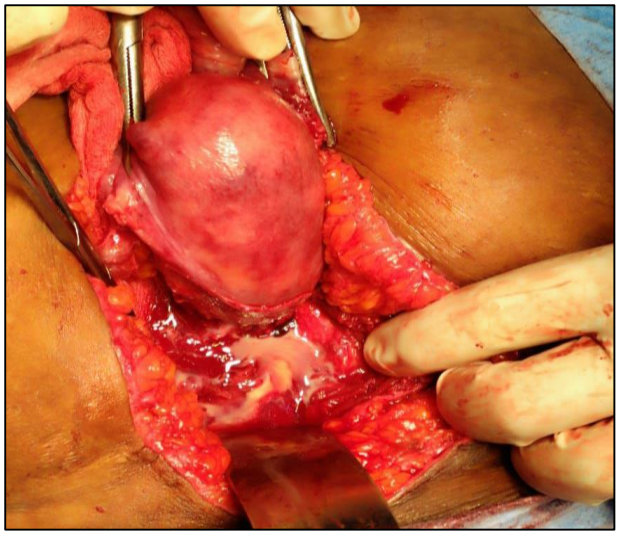

Figure 4: Intra operative picture showing pus draining from the ruptured.

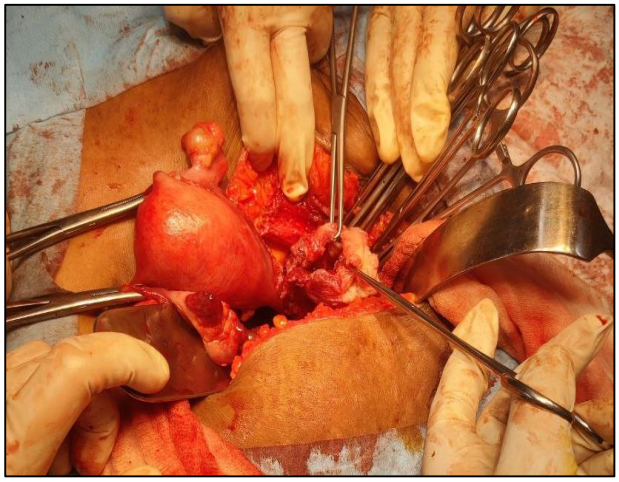

Figure 5: Edges of isthmocele.

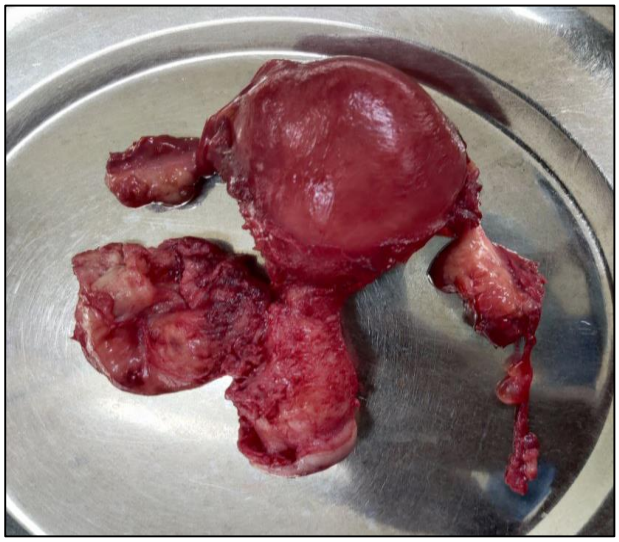

Figure 6: Intact isthmocele removed along with TAH + BSO

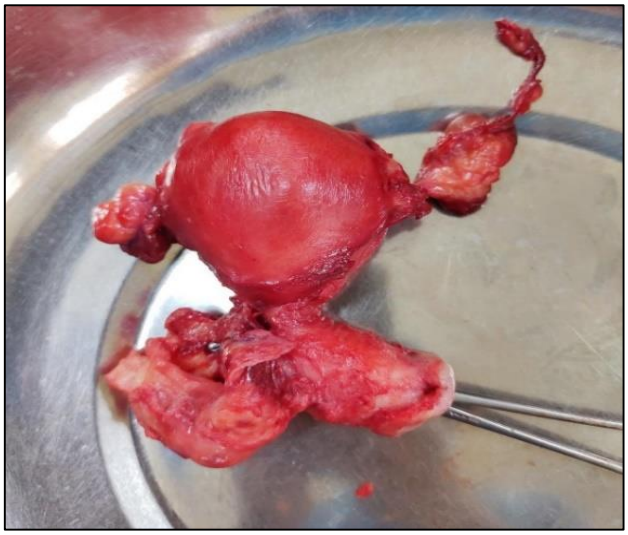

Figure 7: Probe communicating through the cervical canal into isthmocele.

DISCUSSION

Uterine isthmocele is a pouch like diverticulum that forms because of myometrial thinning defect at the site of Caesarean scar in the anterior wall of lower uterine segment. The entity "isthmocele" was first described by Hugh Morris in 1995 who studied a series of 51 hysterectomy specimens and identified the pathological changes at the CS scar.[3] Although in majority of cases isthmocele remains asymptomatic, the most frequently observed symptoms of isthmocele are intermenstrual spotting, dysmenorrhea, dyspareunia, and chronic pelvic pain owing to collection of blood in the pouch. There is an increase in obstetrical sequelae, such as secondary infertility as the collection may make it hostile for the sperm, increased chance of CS scar ectopic pregnancy, placenta accreta spectrum, scar dehiscence or even scar rupture due to poor healing of CS incision.[4] Hence a high index of suspicion is needed in patients with previous CS to evaluate for the presence of the defect in subsequent pregnancies.

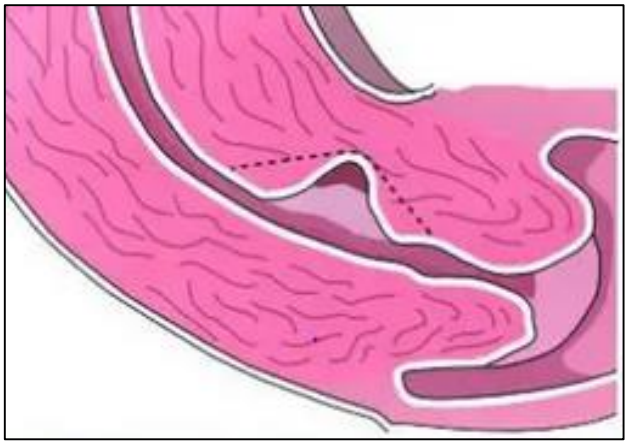

Figure 8: Uterine isthmocele.[3]

The following are some of the most common factors contributing to isthomocele formation. Multiple cesarean sections (CS), inadequate suturing, lower position of CS incision, incomplete closure of the uterine wall due to a single-layer endometrial-saving closure technique or use of locking sutures; and surgical interventions that promote adhesion formation (e.g., non- closure of the peritoneum, inadequate haemostasis, visible sutures, etc.). Patients with a CS performed during active labour with advanced cervical effacement (in case of a cervical dilatation > 5 cm or a labour duration of >5 hours) can also predispose to this condition. Inadequate haemostasis and non-closure of peritoneum promoting adhesion formation, retroflexed uterus and a genetic predisposition also contribute to isthmocele.[5]

Isthmocele can be classified as a small or large defect depending on the wall thickness of the residual myometrium over the niche. According to Marotta et al an isthmocele can be classified as a large defect if the residual myometrium is less than 3 mm and as a small defect if the residual myometrium is more than 3 mm.[6]

Conventionally it is being termed as 'large' if the size of the defect involves more than 50% of the myometrial thickness. For diagnosing an isthmocele, several imaging modalities may be used to assess the integrity of the uterine wall. Transvaginal sonography, Saline infusion Sono hysterogram (SIS), Hysterosalpingogram (HSG), Hysteroscopy and MRI are various modalities to make the diagnosis. However, TVS and SIS are the specific, sensitive and cost-effective methods to diagnose isthmocele. A "filling defect," anechoic and triangular shape under the bladder recess, in the region between the uterine body and the cervix is the typical site identified by TVS or SIS. MRI also shows similar findings that are best depicted on T2 images and are used to accurately measure the defect in 3 planes preoperatively.[7]

Treatment includes medical or surgical management depending on the presence of symptoms, desire for childbearing secondary infertility, site and size of defect. Studies have evaluated the effectiveness of combined hormonal therapy in patients with HMB or intermenstrual spotting after caesarean section in which combined oral contraceptive pills containing 0.5 mg of norgestrel and 0.05 mg of ethinyl estradiol were used. But it was observed that if medical management fails or if the defect is large, surgical management is a better option.[8]

Surgical management includes minimally invasive procedures such as hysteroscopic, laparoscopic, transvaginal approaches or even laparotomy. Choice of open v/s minimally invasive surgery depends on the site and size of defect. Surgical management is opted even in asymptomatic patients if future pregnancy is planned. For small defects, hysteroscopy resection has been reported as a safe, fast, and efficient method in controlling symptoms for the patients who do not desire fertility.[9,10] For larger defects, hysteroscopy has been associated with an increased risk of uterine perforation and bladder injuries. Hence combined hysteroscopic and laparoscopic repair has advantages of both, which achieves best repair especially for defects >3 mm. It reduces complications and is best for patients desiring future pregnancy.[11] For large defect and family completed patients, open laparotomy and surgical resection is the best management.[12,13] Concurrent hysterectomy may have to be planned if associated gynaecological problems like fibroids, adenomyosis, AUB or chronic PID are present.

The use of surgery for the treatment of symptomatic isthmocele is found to improve the bleeding symptoms in more than 80% of patients. However, evidences are lacking regarding the role of surgery for the purpose of improving fertility or reducing the risk of obstetric complications in women with asymptomatic isthmocele.[14]

An abscess developing in an isthmocele long after a CS is considered very rare (only 2 cases were reported as per a study done by department of gynaecology-obstetrics, university hospital of Geneva.[15]

Our patient has H/o of 2 Prev caesareans in view of failed induction and prolonged labour. She had irregular cycles and dyspareunia since second caesarean section, and was severely anaemic. MRI and USS confirmed the diagnosis of isthmocele. As it was a large defect in a perimenopausal age group, and patient was symptomatic with HMB leading to severe anaemia, proceeded with Total abdominal hysterectomy (TAH) and bilateral salpingo-oophorectomy (BSO).

CONCLUSION

Uterine isthmocele is a late and rare complication of caesarean deliveries. It is a frequently overlooked consequence of caesarean scars. As the number of C-section are on the increase, it is important to be aware of this entity and its various presentations to make an early diagnosis. Awareness about this condition and its imaging features is essential to make a prompt diagnosis. Surgical management is indicated in all symptomatic patients and even asymptomatic patients desiring for future pregnancies. Isthmocele presenting as chronic abscess is a very rare presentation and has been reported in only 3 cases in Switzerland till 2016. Such presentations may be easily confused with pyometra or fibroid degeneration and need prompt evaluation and management might be delayed. Hence presenting this rare entity of chronic abscess in isthmocele in a perimenopausal women.

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REFERENCES

1. 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. Human Reproduction. 2015;30(12):2695-702.
2. Kremer TG, Ghiorzi IB, Dibi RP. Isthmocele: an overview of diagnosis and treatment. Revista da Associação Médica Brasileira. 2019;65:714-21.
3. Morris H. Surgical pathology of the lower uterine segment caesarean section scar: is the scar a source of clinical symptoms? Int J Gynecological Pathol. 1995;14(1):16-20.
4. Raimondo G, Grifone G, Raimondo D, Seracchioli R, Scambia G et al. Hysteroscopic treatment of symptomatic cesarean- induced isthmocele: a prospective study. J Minim Invasive Gynecol. 2015;22(2):297-301.
5. Urman B, Arslan T, Aksu S, Taskiran C. Laparoscopic Repair of Cesarean Scar Defect "Isthmocele". J Minim Invasive Gynecol. 2016;23(6): 857-8.
6. Marotta ML, Donnez J, Squifflet J, Jadoul P, Darii N, Donnez O. Laparoscopic repair of post-cesarean section uterine scar defects diagnosed in nonpregnant women. J Minimally Invasive Gynecol. 2013;20(3):386-91.
7. Rupa R, Kushvaha S, Venkatesh K. Uterine isthmocele-a frequently overlooked complication of cesarean sections. Indian J Radiol maging. 2021;31(03):601-4.
8. Tahara M, Shimizu T, Shimoura H. Preliminary report of treatment with oral contraceptive pills for intermenstrual vaginal bleeding secondary to a cesarean section scar. Fertil Steril. 2006;86(2):477-9.
9. Abacjew-Chmylko A, Wydra DG, Olszewska H. Hysteroscopy in the treatment of uterine cesarean section scar diverticulum: a systematic review. Adv Med Sci. 2017;62(2):230-9.
10. Gubbini G, Centini G, Nascetti D, Marra E, Moncini I. Surgical hysteroscopic treatment of cesarean-induced isthmocele in restoring fertility: prospective study. J Minim Invasive Gynecol. 2011;18(2):234-7.
11. Urman B, Arslan T, Aksu S, Taskiran C. Laparoscopic repair of cesarean scar defect “isthmocele”. J Minimally Invasive Gynecol. 2016;23(6):857-8.
12. Florio P, Filippeschi M, Moncini I, Marra E, Franchini M, Gubbini G. Hysteroscopic treatment of the cesarean-induced isthmocele in restoring infertility. Curr Opinion Obstetr Gynecol. 2012;24(3):180-6.
13. Kremer TG, Ghiorzi IB, Dibi RP. Isthmocele: an overview of diagnosis and treatment. Revista da Associação Médica Brasileira. 2019;65:714-21.
14. Vitale SG, Ludwin A, Vilos GA, Török P, Tesarik J, Vitagliano A et al. From hysteroscopy to laparoendoscopic surgery: what is the best surgical approach for symptomatic isthmocele? A systematic review and meta-analysis. Archives of gynecology and obstetrics. 2020;301:33-52.
15. Boukrid M, Dubuisson J. Conservative management of a scar abscess formed in a cesarean-induced isthmocele. Frontiers Surg. 2016;3:7.

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