Postmenopausal bleeding in a woman with caesarean scar defect: A case report

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ABSTRACT

Background: Caesarean scar defect (CSD) is a complication of caesarean section with implications for abnormal uterine bleeding. Most cases of CSD are discovered incidentally or with the development of menstrual changes. However, CSD may rarely have a longer latency period, resulting in postmenopausal presentation of bleeding and abdominal pain.

Case: A 52-year-old postmenopausal woman presented with acute suprapubic pain and postmenopausal bleeding. Pelvic ultrasonography and magnetic resonance imaging suggested bleeding from a CSD. Her symptoms resolved with expectant management. She subsequently underwent elective hysterectomy and bilateral salpingo-oophorectomy, which provided histological confirmation of a CSD.

Conclusion: Caesarean scar defect should be considered in patients who present with acute abnormal uterine bleeding or pelvic pain with a history of caesarean section, even after menopause.

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1. Introduction

Caesarean scar defect (CSD) occurs by the formation of a uterine diverticulum at the site of a previous caesarean section incision. With increasing caesarean section rates, more attention has been placed on the gynaecological complications of CSD, which include abnormal uterine bleeding (AUB), dysmenorrhoea, chronic pelvic pain, and secondary infertility [1]. Recognised AUB changes from CSD include postmenstrual spotting, intermenstrual bleeding, heavy menstrual bleeding, and postcoital bleeding [2]. We discuss a case of CSD with a delayed presentation of postmenopausal bleeding and abdominal pain, for which a hysterectomy and bilateral salpingo-oophorectomy were performed.

2. Case Presentation

A 52-year-old Chinese woman presented to the emergency department with first presentation of acute and severe suprapubic pain for two days, followed by postmenopausal bleeding with passage of blood clots. There were no urinary or bowel symptoms, nausea, loss of appetite, or fever. She denied any recent fall or abdominal trauma. The pain had started spontaneously without any precipitating factors.

Her medical history included hypothyroidism managed with levothyroxine. Her obstetric history included a full-term normal vaginal delivery in her mid-twenties and an uncomplicated elective lower segment caesarean section (LSCS) via Pfannenstiel incision in her mid-thirties. She denied other abdominal or uterine manipulation surgeries. She underwent menopause at 51 years of age, and previously had regular menses with normal flow and no intermenstrual bleeding.

On physical examination, suprapubic tenderness without signs of acute abdomen was noted. Speculum examination revealed a normal vagina and cervix with minimal bleeding from the os. On vaginal examination, the uterus was 12 weeks in size and retroverted, with tenderness on deep palpation. No cervical excitation, adnexal masses, or adnexal tenderness was elicited. Digital rectal examination was unremarkable. She was afebrile and her vital signs were stable.

Urine dipstick was positive for red blood cells (1+) without evidence of protein, leucocytes, or nitrites. Complete blood count and electrolytes were normal with a haemoglobin of 12.3 g/dL. Urine analysis and urine culture were unremarkable. A Pap smear done earlier in the year was normal.

She was admitted in view of her pain and started on empirical broad-spectrum intravenous antibiotics.

2.1. Diagnostic Assessment

Pelvic ultrasonography (Fig. 1) revealed a $5.8 \times 3.3 \times 4.7$ cm mass in the retroverted uterus containing fluid-level echoes, confluent with the lower anterior wall of the uterus and endometrial cavity. Uterine volume was $127 \text{ cm}^3$ and endometrial thickness was
4.1 mm. There was no fluid in the pouch of Douglas and no other masses seen in the pelvis. The left ovary appeared normal, while the right ovary was not visualized. A small intramural fibroid of 3.1 cm$^3$ was noted in the posterior lower uterine wall. The radiological impression was of a lower segment caesarean section scar defect with clots eluding through the scar defect.

A pelvic magnetic resonance imaging (MRI) scan (Fig. 2) showed marked thinning of the anterior uterine wall with corresponding dilatation and bulging of the endometrial cavity, likely related to previous LSCS scar defect, without definite breach of the anterior uterine wall. Fluid layering and T1W/T2W intermediate signals were seen within the distended endometrial cavity, likely related to haemorrhagic products. No gross uterine mass was seen.

As her uterus was acutely retroverted and the myometrium covering the protruding segment appeared very thin, endometrial sampling to exclude malignancy as a cause of postmenopausal bleeding was not attempted to avoid an inadvertent uterine perforation. She was discharged after two days as the vaginal bleeding had resolved.

### 2.2. Therapeutic Intervention

A total hysterectomy was recommended to avoid further episodes of pain and bleeding, the theoretical risk of rupture of the CSD, and to achieve a histological diagnosis and exclude endometrial or cervical cancer. A total laparoscopic hysterectomy and bilateral salpingo-oophorectomy were performed two weeks later.

Intraoperatively, the bladder was noted to be adherent to the lower uterine segment (LUS). It was dissected downwards to reveal a grossly normal-appearing anterior uterine surface. Both fallopian tubes and ovaries appeared normal, as was the rest of the intraperitoneal survey.

### 2.3. Outcome

The specimen was examined after excision (Fig. 3). A probe placed in the uterine cavity via the cervix revealed a diverticulum with an area of thinned myometrium forming a repressed cavity.

Histological examination of the uterus revealed a 1.4 × 1.1 cm area of wall thinning at the anterior aspect of the LUS, measuring 0.6 cm in thickness (Fig. 4). Microscopic examination of this area showed myometrial wall replaced by hyalinization and fibrosis with areas of...
haemorrhage, consistent with a diagnosis of CSD. The serosal surface showed fibrous adhesions, in keeping with the site of a previous surgical scar. The endometrium showed basal-type inactive endometrium, without evidence of hyperplasia, chronic endometritis, or malignancy.

She had an uneventful postoperative recovery and was discharged three days later.

3. Discussion

A CSD, otherwise known as isthmocoele or uterine niche, is any indentation representing myometrial discontinuity in the uterine wall with the base communicating to the uterine cavity, at the site of a previous caesarean section scar [3].

The delayed presentation of CSD is unique as the patient had no menstrual irregularities or abdominal pain before menopause. As she did not conceive after the caesarean section, there were no opportunities for obstetric presentation of CSD, which include caesarean scar ectopic pregnancy, placenta accreta or praevia, scar dehiscence, or uterine rupture [3]. A literature review on the different presentations of CSD did not identify any postmenopausal patients at the time of diagnosis with such a long latency (18 years).

We postulate that the patient had a longstanding CSD which presented when she had an occult postmenopausal bleed. This likely resulted in accumulated blood which stretched the diverticulum, causing sudden lower abdominal pain. The CSD contained fluid with low-level echoes on radiological examination, suggesting recent bleeding at time of presentation, rather than accumulated old blood. The cause of postmenopausal bleeding is unknown, but there was no malignancy on histological examination. We postulate that the bleeding was due to an occult menstrual bleed as her last period was only slightly over a year previously.

Proven risk factors for CSD formation include uterine retroversion and multiple caesarean sections [3]; the former was present in this case. Mechanical traction from retroversion has been suggested to result in greater wound tension and impaired scar perfusion and healing. However, uterine retroversion may also result from CSD due to the lack of support of the corpus by the incomplete uterine wall closure [4]. Other possible risk factors include labour before caesarean delivery and surgical technique used in uterine closure [5].

Radiologically, CSDs demonstrate at least one of four key sono-graphic findings: 1) a wedge defect with a depth of at least 1 mm and an indentation of the myometrium of at least 2 mm in the uterine isthmus at the caesarean section scar site [6], 2) inward scar protrusion, 3) outward protrusion and haematoma, or 4) scar retraction [7]. Rarely, a cystic mass may bulge anteriorly under the bladder. This typically contains low-level echoes consistent with unclotted menstrual blood [8], similar to the ultrasonography images in this case.

The MRI scan also enables evaluation of the thickness of the LUS, depth of the CSD, and the contents of the endometrial and niche cavities. It further aids in the exclusion of other associated pathologies such as adenomyosis or adnexal, uterine, or pelvic diseases [6].

Pathological examination of the uterus showed a thinned uterine wall replaced by fibrosis and areas of haemorrhage, which was consistent with the diagnosis [9,10]. A series of pathological changes have been described in CSDs, including distortion and widening of the LUS, compensatory “overhang” of congested endometrium above the scar recess [11], polyp formation conforming to the contours of the scar recess, moderate to marked lymphocytic infiltration, residual suture material with foreign body giant cell reaction, capillary dilatation, fragmentation and breakdown of the endometrium of the scar, and iatrogenic adenomyosis confined to the scar [9]. It is also postulated that the fibrotic scar tissue hinders normal endometrial development and synchronization with the surrounding endometrial lining, as secretory endometrium was found on histology in 52.5% of cases of hysteroscopic resection of CSD scheduled during the early follicular phase [12].

4. Conclusion

This is the first reported case of a CSD presenting as postmenopausal bleeding many years after the caesarean section. With the increasing incidence of caesarean sections worldwide, clinicians should be mindful of this possibility when encountering patients who present with AUB or pelvic pain, even after menopause.

Contributors

All authors contributed equally to the preparation of this case report and saw and approved the final manuscript.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.
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Patient Consent

The patient described in this case study provided written informed consent.

Provenance and Peer Review

This case report was peer reviewed.

References

[1] N. Schepker, G.-J. Garcia-Rocha, F. von Versen-Höynck, P. Hillemanns, C. Schippert, Clinical diagnosis and therapy of uterine scar defects after caesarean section in non-pregnant women, Arch. Gynecol. Obstet. 291 (2015) 1417–1423, https://doi.org/10.1007/s00404-014-3582-0.

[2] Y.-Y. Chen, C.-C. Tsai, F.-T. Kung, K.-C. Lan, Y.-C. Ou, Association between hysteroscopic findings of previous caesarean delivery scar defects and abnormal uterine bleeding, Taiwanese J. Obstet. Gynecol. 58 (2019) 541–544, https://doi.org/10.1016/j.tjog.2019.05.020.

[3] T. Kremer, I. Ghiorzi, R. Dibi, Isthmocele: an overview of diagnosis and treatment, Rev. Assoc. Méd. Bras. 65 (2019) 714–721, https://doi.org/10.1590/1806-9282.65.5714.

[4] J. Park, M. Kim, H. Lee, Y. Gen, M. Kim, Risk factors for Korean women to develop an isthmocele after a cesarean section, BMC Pregnancy Childbirth 18 (2018), 162. https://doi.org/10.1186/s12884-018-1821-2.

[5] A.M. Tower, G.N. Frishman, Cesarean scar defects: an underrecognized cause of abnormal uterine bleeding and other gynecologic complications, J. Minim. Invasive Gynecol. 20 (2013) 562–572, https://doi.org/10.1016/j.jmig.2013.03.008.

[6] A. Setubal, et al., Treatment for uterine Isthmocele, a Pouchlike defect at the site of a Cesarean section scar, J. Minim. Invasive Gynecol. 25 (2018) 38–46, https://doi.org/10.1016/j.jmig.2017.09.022.

[7] H. Chen, S. Chen, F. Hsieh, Observation of cesarean section scar by transvaginal ultrasonography, Ultrasound Med. Biol. 16 (1990) 443–447, https://doi.org/10.1016/0301-5629(90)90166-A.

[8] B.R. Benacerraf, S.R. Goldstein, Y.S. Groszmann, Cesarean scar defect, Gynecologic Ultrasound: A Problem-Based Approach, Elsevier/Saunders 2014, pp. 39–42.

[9] G. Gubbini, P. Casadio, E. Marra, Resectoscopic correction of the “Isthmocele” in women with postmenstrual abnormal uterine bleeding and secondary infertility, J. Minimal. Invasive Gynecol. 15 (2008) 172–175, https://doi.org/10.1016/j.jmig.2007.10.004.

[10] B. Lotti, et al., Cesarean-induced Isthmocele, Surgical hysteroscopic treatment and reproductive outcome, Fertil. Steril. 100 (2013) 5397, https://doi.org/10.1016/j.fertnstert.2013.07.681.

[11] A.A.A. Ewies, U. Zanetto, Cesarean section scar causes myometrial hypertrophy with subsequent heavy menstrual flow and dysmenorrhea, Med. Hypotheses 108 (2017) 54–56, https://doi.org/10.1016/j.mehy.2017.08.006.

[12] N. Schepker, G.-J. Garcia-Rocha, F. von Versen-Höynck, P. Hillemanns, C. Schippert, Clinical diagnosis and therapy of uterine scar defects after caesarean section in non-pregnant women, Arch. Gynecol. Obstet. 291 (2015) 1417–1423, https://doi.org/10.1007/s00404-014-3582-0.