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Scar Endometriosis Complicated with Utero-Cutaneous Fistula: Case Report

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Abstract

Background: *Scar endometriosis is a rare complication of cesarean delivery, and its progression to a utero-cutaneous fistula (UCF) is exceptionally uncommon. This paper describes a rare type of fistula after a long time of cesarean section, and increases awareness regarding this rare condition, which is often misdiagnosed.*

Case presentation: *We report the case of a 35-year-old multiparous woman with a history of three cesarean sections who presented with cyclic bleeding from a poorly healed Pfannenstiel scar and severe dysmenorrhea. Clinical examination revealed two discolored subcutaneous masses along the previous incision site. Ultrasonography and CT imaging demonstrated subcutaneous lesions adherent to the anterior uterine wall, while pelvic MRI confirmed a hyperintense tract establishing communication between the uterine cavity and the abdominal wall. The patient underwent wide surgical excision of the endometriotic masses with removal of the fistulous tract, reconstruction of the uterine wall, and placement of a mesh to repair the abdominal wall defect. Histopathology confirmed endometriosis and utero-cutaneous fistula. Postoperative recovery was uneventful, and symptoms resolved completely.*

Conclusion*: This case highlights the importance of considering scar endometriosis in women presenting with cyclic symptoms at previous cesarean sites and emphasizes the role of MRI in diagnosing rare complications such as UCF. Complete surgical excision remains the definitive and effective treatment.*

Key words: scar endometriosis, utero-cutaneous fistula, previous cesarean sections, cyclic cesarean scar bleeding.

Abbreviations: CSE = Cesarean scar endometriosis, AWE = Abdominal wall endometriosis, MRI = Magnetic resonance imaging, GnRH = Gonadotropin-releasing hormone, CSE = Cesarean scar endometriosis, UCF = Utero-cutaneous fistula, IUCD = intrauterine contraceptive device, CS = Cesarean section.

Introduction

The occurrence of scar endometriosis with utero-cutaneous fistula represents an exceptionally rare complication following cesarean delivery. Endometriosis itself is a hormone-dependent gynecological disorder that affects 2–10% of all women in their reproductive age (1). First recognized by Rokitansky in 1860 (2). The hallmark of this illness is characterized by the ectopic presence of endometrial glands and/or stroma outside the endometrium, typically accompanied by varying degrees of an inflammatory process (3). Although the pelvic cavity, particularly the ovaries, fallopian tubes, and uterosacral ligaments is the most prevalent site of disease, while the remote areas that may be affected outside the pelvis are the abdominal wall, episiotomy scars, urinary and gastrointestinal tract, nasal mucosa, and even the thorax (4). Abdominal wall EM (AWE) is the rarest form of EM (5), which is defined as the presence of endometrial tissue between the parietal peritoneum and skin. Anatomically, classified as superficial (Cutaneous/ subcutaneous), intermediate (invading fascia), or deep (involving muscle fibers) (6). Among these forms, CSE is the most common and frequently encountered (7). CSE often arises after obstetric and gynecologic surgical procedures, especially caesarean delivery, when the endometrium is involved, and has been reported even when the endometrium was not breached (8). According to Nominato et al, cesarean delivery markedly raises the likelihood of developing AWE (9). The incidence rates range from 0.03% to 3.5% of postcesarean patients (10). Regarding the pathophysiology of scar endometriosis, the exact cause remains debated, but the most widely accepted theories are unintentionally seeded of endometrial cells into the incision during cesarean section (11). These cells may then survive, respond to cyclic hormonal stimulation, and potentially induce metaplastic changes in adjacent tissues, resulting in CSE. Additionally, the lymphatic or vascular pathways may be responsible for how the endometrial tissue may reach the wound (9). CSE commonly appears in or adjacent to cesarean scars (12) and about 80% of cases occur at the lateral edges of the scar (13). Over time, the repeated periodic menstrual bleeding combined with chronic Inflammation and cellular proliferation of the ectopic endometrial tissue implants may lead to tissue necrosis formation, establishing an abnormal tract that connects the endometrial tissue in the scar to the uterine cavity and may lead to utero-cutaneous fistula formation. In general, the fistula is a communication between two surfaces lined with epithelium. This abnormal connection occurs after traumas, infections, or other injuries (14). The most common uterine fistulae are uterovesical or utero-colonic due to injuries or infection after the surgery (15). In contrast, utero-cutaneous fistulas are scarcely reported and remain a highly unusual complication.

Case report

A thirty-five-year-old divorced multiparous woman presented to the out-patient gynecological clinic as a known case of diabetes controlled with treatment, and her chief complaints of bleeding per cesarean scar, as shown in Figure 1a. Her obstetrical history was three previous lower-segment cesarean sections with one abortion, the last cesarean section

done eight years back, and otherwise unremarkable. The postoperative period was uneventful except that the second cesarean section was complicated by wound infection for more than one month, and she received treatment at the outpatient clinic. Her gynecological history revealed that the menstrual cycle was regular every 28 ± 2 day, with a flow of 5-6 days. After seven months of the last cesarean delivery, the patient presented with pain at the left edge of the operation site and severe dysmenorrhea interfering with daily activity, relieved with analgesia. For the past three years, she had bleeding per cesarean scar, which was linked to an increase in the severity of the pain at the operation site during the menstrual cycle; however, there were no complaints of pain or discharge from the operation site in between menstrual cycles. Her general examination was within normal limits, a part of pallor was evident, and her vital signs were stable. Abdominal examination revealed a cutaneous retraction and poorly healed scar with a brownish, bluish discolored non-mobile mass at two points of the abdominal wall, the first point at the upper border of the right side of the Pfannenstiel caesarean scar, and the second point at the left extreme side of the Pfannenstiel caesarean scar Figure 1b, with slight tenderness, firm consistency, and no discharging blood during pressure over the lower abdomen, vaginal examination by the speculum was normal.

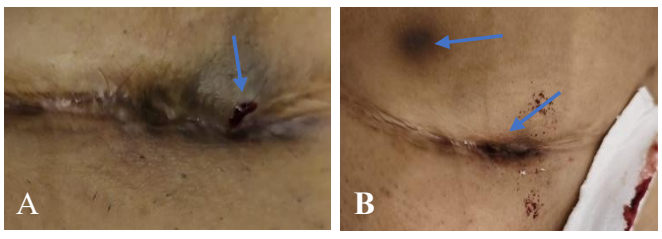

Figure 1: (A) Shows bleeding from the poorly healed cesarean scar/fistula with skin retraction above the node during the menstrual cycle.
(B) Shows a brownish, bluish discolored nonmobile mass at two points of the abdominal wall.

Pelvic ultrasonography was performed, showing one ill-defined, big hypoechogenic mass in the subcutaneous layer on the left lateral side of the Pfannenstiel caesarean scar, as shown in Figure 2, and one well-defined, small mass in the subcutaneous layer on the upper border of the right side of the scar.

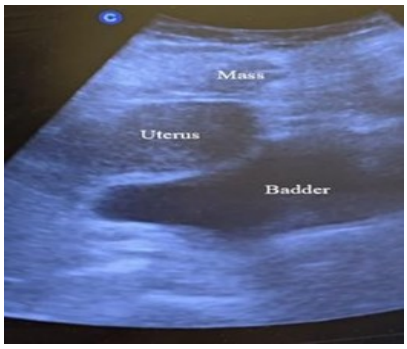

Figure 2: The transabdominal pelvic ultrasound showing an irregular hypoechoic solid mass into the subcutaneous tissues.

CT scan revealed adhesion between the anterior wall of the uterus and subcutaneous tissue of anterior abdominal wall with altered tissue near the incision site and an ill-defined mass involving LT lower rectus abdominus muscle related to the previously cesarean scar, as shown in Figure 3.

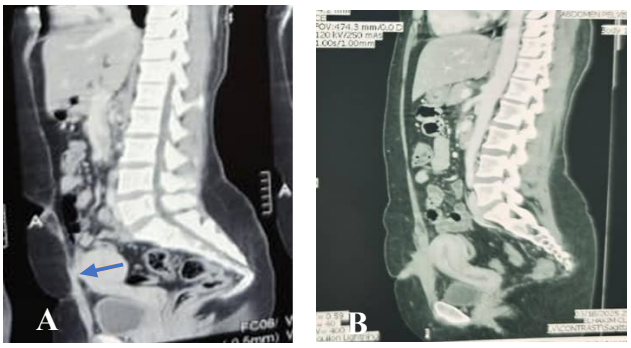

Figure 3: On computed tomography scan, the sagittal plane shows the anterior wall of the uterus adherent to the subcutaneous tissue of the anterior abdominal wall near the scar, as in photo A, extends through the abdominal wall up to the scar site as seen in photo B

Pelvic MRI scan showing the presence of a hyperintense fistula that communicates between the anterior abdominal wall and the uterine cavity, as shown in Figure 4.

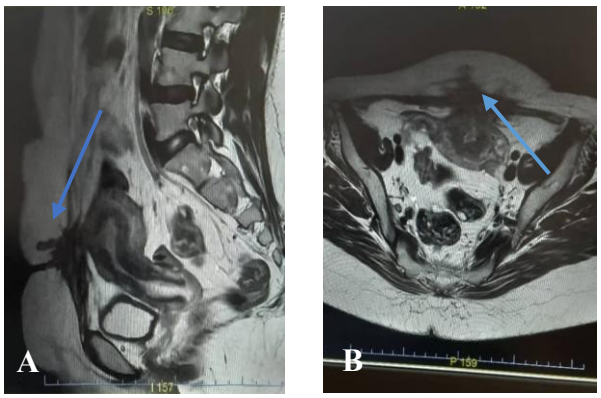

Figure 4: Magnetic resonance imaging, (A) the sagittal and (B) axial views that the blue arrow shows a fistula that communicates between the anterior abdominal wall and the uterine cavity

The provisional diagnosis is cesarean scar endometriosis with UCF was made, and planned for surgical excision. All hematological investigations were within normal limits. Apart from the hemoglobin being 7.9 g/dl, three units of blood transfusions were received, and the tumor marker was within normal limits. Patient underwent laparotomy, the operation was done by a gynecologist and a general surgeon, an elliptical incision was made around the previous cesarean scar to create healthy skin margins, she had a wide local excision of the endometriotic scar, the intraoperative finding includes a large fibrotic nodule extending inferiorly to the symphysis pubis, and there is moderate involvement of the anterior rectus sheath. The anterior uterine wall obliterated with the mass and the uterine fundus adherent to small intestine loop which is released, and the entire lesion were dissected gently from uterine wall, wide-margin surgical excision of the both small and large mass was done with estimated size approximately about 6 cm as shown in Figure 5A, there is gross evidence of endometriotic seedlings at the uterosacral ligament, and they were cauterized, Figure 6 shows a small hole is noted in the center of anterior uterine wall. Uterine incision was done, and the edges of the fistula and necrotic tissue were removed. Refreshment of the edges was performed, after which the endometrial cavity was clearly visible. The cavity was effectively obliterated with stitches, Vicryl number one, and the rest of the uterus was repaired in two layers. Hemostasis was secured, and the sheath was reconstructed later. An intraoperative mesh (27*25) was inserted to repair a defect caused by the resection as shown in Figure 7. Two drains were inserted, and the anterior abdominal wall was closed. The entire lesion is excised and cut open to demonstrate the endometriosis, which is present inside, and typically seen as brown and black spots, as shown in Figure 5B. The sample were sent to the pathology laboratory for definitive histological examination. Postoperatively, the patient received antibiotics and analgesia with dressing, the patient was discharged on the sixth day, and stitches were removed on postoperative day ten; the stitch line has healed without any complications, as shown in Figure 8. The patient is in follow-up, her first cycle after the operation lasted for 5 days without dysmenorrhea and the subsequent visits were also uneventful. Histopathological findings confirmed the diagnosis of scar endometriosis and utero-cutaneous fistula.

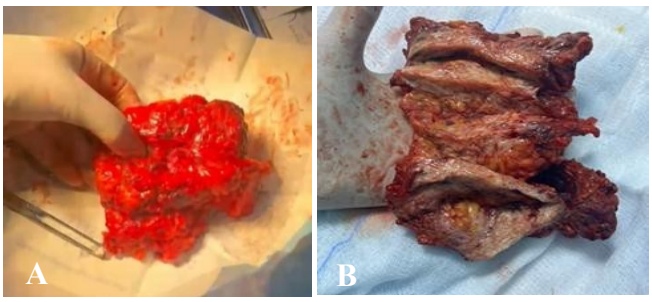

Figure 5: Gross appearance of a specimen (A) This 6 cm, fibrotic nodule excised from the sub-

cutaneous layer of a patient, (B) Cut section of the excised mass revealed a dark red endometriotic spots

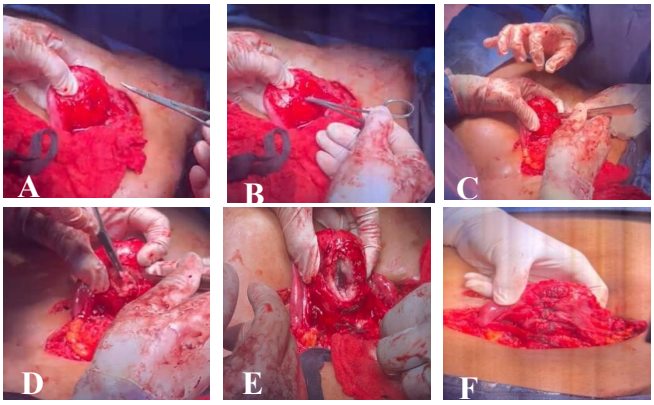

Figure 6: Intraoperative picture showing (A) a hole is noted in the center of the anterior uterine wall, (B) a small artery inserted inside the hole, (C&D) uterine incision around the fistula and removal of the necrotic tissues, (E) exposed endometrial cavity, and (F) the uterine wall after repair

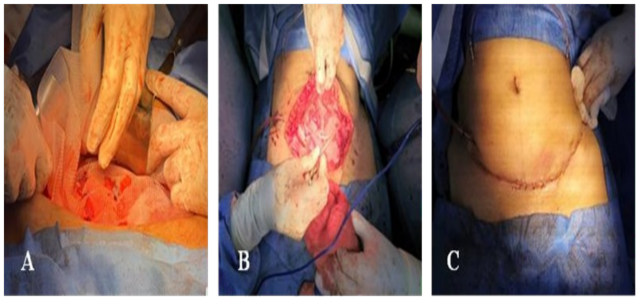

Figure 7: intraoperative picture showing (A) mesh insertion, (B) two drains were inserted, and (C) closure of the anterior abdominal wall.

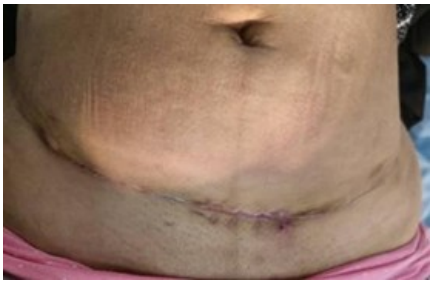

Figure 8. Healed laparotomy scar after excision of the endometrioma.

Discussion

The coexistence of CSE and UCF is extremely rare; and can pose a clinical challenge, as both conditions often present with distressing symptoms during menstruation. Endometriosis typically manifests as cyclic pain and dark discoloration of the skin, with or without periodic bleeding, whereas UCF usually presents primarily with cyclical bleeding (10)(14)(16). The relationship between the two entities is complex, while endometriosis is an uncommon cause of cutaneous fistula, but there are cases of endometriosis developing inside a utero-cutaneous fistula that have been reported (17)(16). In the index case, prior wound infection and dehiscence likely allowed direct contact between the uterus and abdominal wall, potentially contributing to fistula formation. Additionally, inadvertent inclusion of a sub-involuted uterus in the secondary wound closure stitch may have played a role (18). Other predisposing factors include retained placental tissue, multiple prior abdominal surgeries, prolonged drain use, incomplete closure of the uterine incision, and wound dehiscence. Rare causes of UCF may include Actinomyces infection or migration of intrauterine contraceptive devices (IUCDs (19). Although clinical diagnosis of scar endometriosis is challenging, characteristic features such as cyclic pelvic pain and localized swelling can aid in detection (20). Cyclic symptoms such as bleeding or discharge from the surgical scar during menstruation are not always seen, but if present, are pathognomonic for scar endometriosis (21) as seen in our patient, the symptoms appear after eight years of the last cesarean section, consistent with previous studies, which demonstrated that the typical time before symptoms appear in scar endometriosis ranges from three months to eighteen years (22). Diagnostic modalities for scar endometriosis include ultrasonography, computed tomography (CT), magnetic resonance imaging (MRI), and Doppler sonography. Ultrasonography is often the

first-line investigation, while CT and MRI are useful in excluding alternative diagnoses such as lipoma, hernia, or neoplasm (22). Tumor markers, including CA19-9 and CA125, may be normal or elevated, but definitive diagnosis requires histopathological examination following surgical excision (23). MRI demonstrates high sensitivity (90–92%) and specificity (91–98%) for detecting scar endometriosis (24), and is particularly valuable for preoperative mapping of disease extent (25). Nevertheless, the absence of an imaging technique with 100% sensitivity does not rule out endometriosis (26), and histopathological confirmation remains essential (27)(10). The provisional diagnosis of CSE with UCF was based on the patient's history of multiple cesarean sections, one complicated by wound infection, along with clinical and imaging findings, and surgical excision was planned. Nonetheless, the most successful treatment for scar endometriosis is still surgical excision, which is both diagnostic and curative. Offiong et al.(28) recommend cannulation of the fistula during surgery to guide dissection; this was not feasible due to the small size of the fistulous opening, which was only evident during menstruation; hence, we had to rely on tissue planes and tissue texture to guide our dissection. Wide surgical excision with clear margins of at least 1 cm is considered both diagnostic and curative and minimizes the risk of recurrence and rare malignant transformation into endometrioid or clear cell carcinoma, which occurs in approximately 1% of repeated cases (29). Pas et al. (30) suggested that it is crucial to use mesh to restore significant post-excisional deficits in women who plan to become pregnant . Postoperative hormonal therapy, including danazol, GnRH agonists, or progesterone, has been suggested to reduce recurrence, but its efficacy remains debated (31). Recurrence rates following surgical excision are generally low. For instance, Uçar et al. reported no recurrence over 12–60 months of follow-up (32), Horton et al. observed a recurrence rate of 4.3% (7), and Zhang and Li reported a rate of 7.8% over an average of 20 ± 16 months (33). Most authors agree that surgical excision is successful in preventing both recurrence and conversion to malignancy, which has been reported in sporadic cases despite being quite uncommon (34)(35).

Conclusion

Cesarean scar endometriosis should be considered in women of reproductive age who have a history of gynecological or obstetric surgery and present with cyclic pain or swelling at their abdominal incision sites. Early recognition is essential to ensure timely diagnosis and management, as untreated scar endometriosis can lead to complications such as the development of a utero-cutaneous fistula. This condition underscores the importance of meticulous surgical technique during cesarean section, including careful hemostasis, thorough irrigation of the incision site before closure, and the use of separate sponges for the uterine cavity and abdominal wall. These measures may help reduce the incidence of scar endometriosis. Increasing awareness among obstetricians and healthcare providers is crucial for prevention and early detection. Further research is warranted to clarify the underlying mechanisms and improve preventative strategies

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the signed consent form is available for review by the Editor-in-Chief of the journal upon request.

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