

# Intrauterine endometrial cyst after low uterine incision

## A case report with literature review

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### Abstract

**Rationale:** During the surgical procedure, endometrial cells can be seeded into the wound edge of the uterine wall, developing into scar endometriosis. Due to the extremely low incidence, estimation of its prevalence is still unavailable. Even rarer might be the scar endometriosis in uterine cavity, to our best knowledge, a situation has not been reported yet.

**Patient concerns:** A 37-year-old woman complained of heavier and prolonged menstruation as well as pelvic pain during menses for more than 4 months. An endometrial cyst in diameter of 6 cm in uterine cavity was revealed by transvaginal ultrasound. Her surgical history was significant for 1 caesarean section and 1 abdominal myomectomy through transverse incision of lower uterine segment.

**Diagnoses:** Space-occupying lesions in uterine cavity, moderate anemia and scar uterus.

**Interventions:** The hysteroscopy was performed and a multilocular cyst full of chocolate-like fluid was removed. Pathological examination confirmed endometrial glands in the removed cyst tissue.

**Outcomes:** During the follow-up visits at 1 and 6 months after surgery, the patient denied any special discomfort. Her postoperative transvaginal ultrasound showed an enlarged uterus with no lesion in uterine cavity. To achieve a better surveillance, a 3-year period of follow-up after surgery at a 6-month interval was suggested.

**Lessons:** Intrauterine endometriosis should be considered in patients of pelvic surgery history with pelvic pain, menstrual disorder, and intrauterine cystic mass.

**Abbreviations:** EMS = endometriosis, GnRH = gonadotropin-releasing hormone, HGB = hemoglobin, TVU = transvaginal ultrasound.

**Keywords:** abdominal myomectomy, caesarean section, endometrial cyst, hysteroscopy, scar endometriosis

## 1. Introduction

The rate of abdominal scar endometriosis following pelvic surgeries has been reported as 0.03% to 1.96% in different studies.<sup>[1–4]</sup> Besides common locations as abdominal subcutaneous tissue or rectus muscles,<sup>[5]</sup> implantation of endometrial tissue may occur at the incision on the uterine wall as well.<sup>[6–9]</sup> However, due to extremely low incidence, its prevalence rate could not be

estimated yet. Here, we report 1 case of scar endometriosis in the form of cystic mass protruding into the uterine cavity after pelvic surgical procedures. To our best knowledge, this is the first case of intrauterine scar endometrial cyst that has been described. Accompanied by rapidly increasing rate of caesarean section in recent years, more scar endometriosis might occur. We aim to provide new information about this rare entity and make contributions to achieving quicker diagnosis and better prevention.

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## 2. Case presentation

A 37-year-old, gravida 1, parity 1, woman complained of heavier and prolonged menstruation for more than 4 months. Despite of a regular cycle of 30 days, her menstrual period was extended to 7 to 8 days and bleeding volume increased to double times (using 10–20 pieces of heaving flow pads), compared with her usual menstrual period (4–5 days), and volume (using 8–10 pieces of heaving flow pads). Vague pain in pelvic and increased pressure in anus was also complained of during menses. The patient denied dysmenorrhea, dyspareunia, and symptoms associated with anemia including fatigue, dizziness, or shortness of breath.

The surgical history was significant for 1 caesarean section in 2007, 1 hysteroscopic myomectomy in 2014, and 1 abdominal myomectomy in 2015. The caesarean section was performed at term through transverse incision of lower uterine segment. The abdominal myomectomy was also performed through transverse incision of lower uterine segment, during which, a submucosal uterus leiomyoma in size of 6 × 5 × 4 cm was removed.

The patient had no other significant medical history, no remarkable family history or review of system. The pelvic and

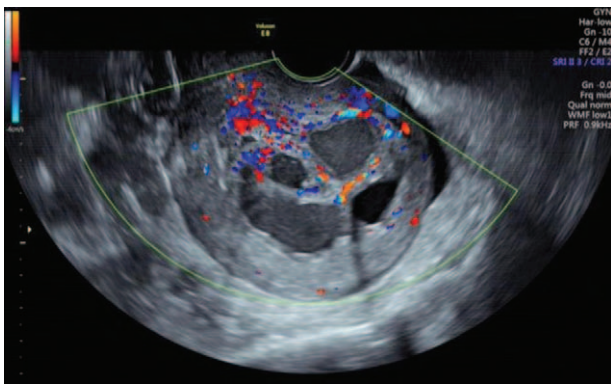


Figure 1. Ultrasonographic view of the cystic mass in the uterine cavity.

rectal examinations revealed an enlarged uterus at size of 2-month gestation. The uterus was mobile and smooth without tenderness. The transvaginal ultrasound revealed a 5.5 × 4.7 × 6.1 cm multilocular cystic mass in the uterus cavity, with irregular fluid sonolucent areas, scattered with bright spot echoes. No lesion was found on bilateral ovaries (Fig. 1). The laboratory tests showed a decreased level of hemoglobin (85 g/L). The primary admitting diagnosis was space-occupying lesions in uterine cavity, among which, submucous myoma was highly suspected. The secondary diagnoses included moderate anemia and scar uterus. Based on the hypermenorrhea and intrauterine abnormal echo by ultrasound, hysteroscopy was suggested for further diagnosis and removal of lesion.

During the hysteroscopic surgery, the uterine cavity was found to be 13 cm deep, occupied by a cystic mass in size of 6 × 5 × 5 cm. The cyst seemed to originate from the front uterine wall and extend to the fundus and right wall. There were multiple divisions in the cyst, full of a large amount of chocolate-like fluid (Fig. 2). The cyst was resected and endometrial ablation was performed around the original area of the lesion. After removing the cyst, the uterine cavity was 10 cm deep with a normal appearance. Pathological examination confirmed endometrial glands and stroma in the removed cyst tissue (Fig. 3). One week after surgery, the patient was discharged in a good condition. The discharge diagnoses were as follow: Endometrial cyst in uterine cavity, intrauterine adhesion, moderate anemia, and scar uterus. The first follow-up was scheduled at 1 month after surgery. The patient denied hypermenorrhea, menostaxis, menstrual cycle disturbance, pelvic pain,



Figure 2. Hysteroscopic view of the cystic mass: Multiple divisions full of chocolate-like fluid.

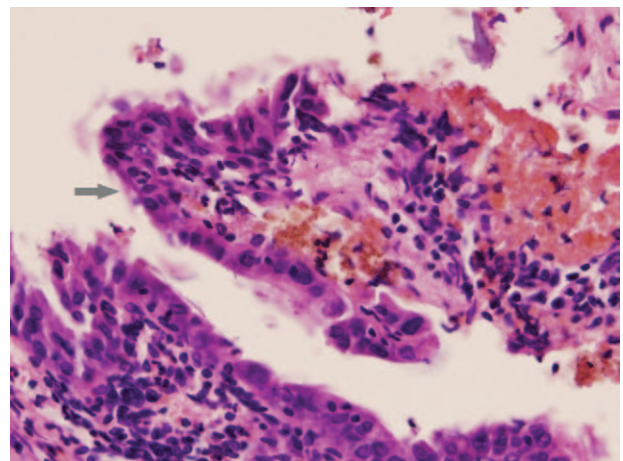


Figure 3. Microscopical view of the series section: The arrow showed endometrial stroma and gland structures in the removed cyst tissues (original magnification ×400).

and other abnormalities. The pelvic examination revealed a mobile and smooth uterus without tenderness. To reduce the risk of recurrence, GnRH agonist or levonorgestrel-releasing intrauterine device was suggested 3 months after surgery. However, the patient refused to follow the advice due to no symptoms. Therefore, a 3-year period of follow-up after surgery at a 6-month interval was suggested to achieve a better surveillance. During the follow-up at 6 months after surgery, the patient denied any special discomfort. Her transvaginal ultrasound showed an enlarged uterus in size of 4.8 × 6.2 × 5.3 cm and no lesion was found in uterine or pelvic cavity (Fig. 4). The relevant data on the episode of care were summarized in Fig. 5. Informed consent was obtained from the patient with guarantees of confidentiality.

### 3. Discussion

Extra-pelvic endometriosis is a rare disease, which may occur at abdominal scar, urinary tract, lungs, intestine, pleural cavity, extremities, and all other sites inclusive.<sup>[10]</sup> Among them, the incidence of abdominal scar endometriosis is especially low but increasing gradually in recent years,<sup>[4,11]</sup> with an incidence rate of 1.08% after hysterotomy<sup>[2]</sup> and 0.03% to 1.96% after caesarean

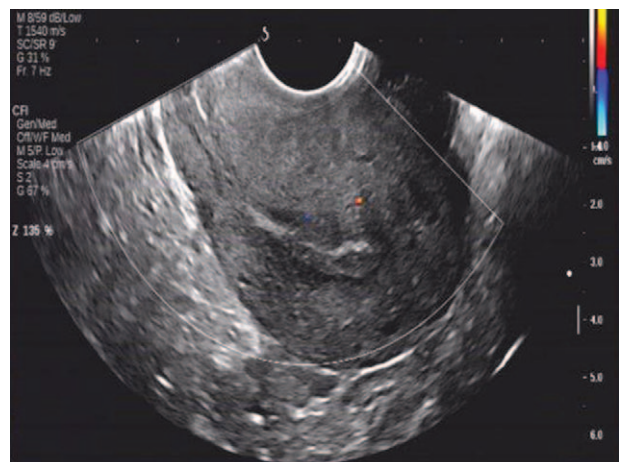


Figure 4. Ultrasonographic view of the uterine cavity at 6 months after surgery.

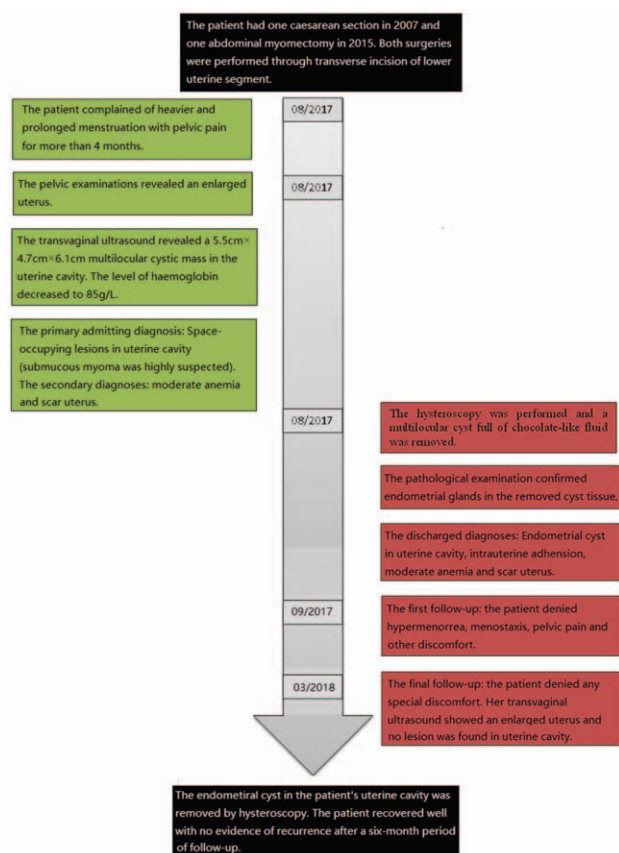


Figure 5. Timeline as a chronological summary of the episode of care.

section,<sup>[1-4]</sup> respectively. Compared with abdominal scar endometriosis, endometriosis in the uterine wall is only reported in few cases,<sup>[6-9]</sup> lacking data of its estimated incidence rate. These reported cases all had a history of caesarean section and the observed endometrial cysts in the scar tissue ranged from 1 to 1.5 cm in diameter.<sup>[6,7]</sup> In our case, a chocolate cyst in diameter of 6 cm appeared in the uterine cavity after abdominal myomectomy for submucosal uterus leiomyoma.

Unlike abdominal scar endometriosis, typically presenting with cyclic abdominal wall pain and palpable mass adjacent to previous surgical scar, the diagnosis of intrauterine scar endometriosis is more challenging.<sup>[12]</sup> The clinical manifestation varies from chronic pelvic pain, dyspareunia to dysmenorrhea, without clear indicators of location of the disease.<sup>[7]</sup> This could partly explain the underestimated occurrence of uterus scar endometriosis. As shown by a retrospective review, endometriosis confined to previous caesarean section scar was observed among 28% hysterectomy specimens.<sup>[13]</sup> In our patient, the major clinical manifestation was excessive and prolonged menstruation accompanied by vague pain in pelvic during menses. Unlike in previous cases, the endometriosis lesion was not confined in the uterine wall, but growing into the uterine cavity as an endometrial cyst of 6 cm in diameter.

The time interval between the latest pelvic surgery and the diagnosis of abdominal scar endometriosis varied from 45 days to 20 years in different studies.<sup>[4,11,12]</sup> For uterus scar endometriosis, the time gap is reported as 6 weeks to 9 years in 3 cases.<sup>[7,8]</sup> In our patient, onset of symptoms started 10 years after caesarean section and 2 years after abdominal myomectomy.

Based on the fact that most cases of scar endometriosis had a history of pelvic surgery, the “iatrogenic direct implantation” has been postulated as the underlying mechanism.<sup>[12]</sup> During the surgical procedure, endometrial cells can be seeded into the wound edge of the subcutaneous tissue or the uterine wall, developing into abdominal scar endometriosis or uterus scar endometriosis, respectively. It has been hypothesized that suboptimal closure of uterine incision or pelvic layers, such as sutures through the decidua, may lead to endometriosis in the scar tissue.<sup>[7,12]</sup> Especially for patients of obesity or with a thin lower uterine segment, their operations are usually more difficult, which results in unsatisfied control of the depth of sutures.<sup>[12]</sup> In addition, wiping the uterus cavity with dry or wet sponges may increase the risk of scar endometriosis via extruding endometrium tissue into the incision site.<sup>[5,7,14]</sup> In our case, the patient had twice surgical procedures penetrating through the entire myometrium to the uterine cavity (caesarean section and abdominal myomectomy). As a result, some endometriosis tissues might be entrapped in the uterine wall during operations. While some ectopic decidual tissue remained stationary, a part of implanted endometrium near mucous layer has developed into a 6 cm endometrial cyst in uterine cavity as described.

The surgical excision of endometrial lesions was the definitive treatment for scar endometriosis. Alternative methods include oral contraceptives, progesterone, danazol, and GnRH agonists.<sup>[12,15]</sup> The recurrence rate of 4.5% to 9.1% is reported after surgical excision, while no data are available on effect of postoperative hormonal suppressive treatment or other medical therapies.<sup>[12,15]</sup> We performed a hysteroscopy for the patient and removed the endometrial cyst. The patient recovered well with no evidence of recurrence after a 6-month period of follow-up.

In conclusion, the incidence of uterus scar endometriosis might be underestimated. Due to increasing rate of caesarean section in recent years, more scar endometriosis might occur as a result. As it is usually difficult to identify this rare disease, our case provides valuable information. Intrauterine endometriosis should be considered in patients with pelvic pain, menstrual disorder, and intrauterine cystic mass after pelvic surgery. Careful suture of uterine incision and avoidance of endometrium wiping during surgical procedure may reduce the risk of later scar endometriosis. Further studies are needed to investigate the factors associated with the recurrence after surgery.

### Author contributions

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