Post-Cesarean Section Abdominal Wall Endometriosis Requiring Surgical Treatment: A Case Series

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The abdominal wall is an uncommon site for endometriosis that occurs in the soft tissues of the wound site after laparotomy. The present study reviewed cases of post-cesarean section abdominal wall endometriosis that were surgically treated at our institution from April 2007 to August 2020. We analyzed data from nine patients who were diagnosed with abdominal wall endometriosis and selected surgery after receiving sufficient explanation of hormone therapy and from patients who reported no improvement in symptoms with hormone therapy. Written consent for case reporting was obtained from all patients. The location of abdominal wall endometriosis was at the wound site in seven cases and outside the wound in two cases. In addition, 7/9 (78%) of post-cesarean cases of abdominal wall endometriosis were related to the cesarean procedure, and 6/9 (67%) of the cases had symptoms related to menstruation. Post-cesarean abdominal endometriosis was associated with pain (7/9; 78%) and a palpable mass (3/9; 33%). Surgical resection did not cause large tissue defects, and artificial repairs such as synthetic mesh were not required for any patient. In conclusion, women with a history of a cesarean section who present with recurrent pain or masses in the wound or abdominal wall that coincide with the menstrual cycle should be closely evaluated for abdominal wall endometriosis. Because surgical treatment often requires postoperative wound reconstruction, collaboration with plastic surgeons and dermatologists is essential. (J Nippon Med Sch 2024; 91: 560-566)

Key words: abdominal wall, cesarean section, endometriosis, surgery, transplants

Introduction

Endometriosis is a disease in which endometrium-like tissue develops outside the endometrium. Although it develops at various sites and can affect any body part, incidence and symptoms vary greatly in relation to the site¹. The abdominal wall is an uncommon site of endometriosis that develops in soft tissues such as subcutaneous, subfascial, or muscle tissue—often at the wound site after laparotomy². The incidence of abdominal wall endometriosis after obstetric and gynecologic surgery is increasing because of the increased number of cesarean sections performed worldwide³. Approximately 80% of cases of abdominal wall endometriosis occur in the cesarean section wound⁴. Additionally, women have developed endometriosis in the umbilical or inguinal region, even without previous surgery⁵. The present study reviewed nine cases of post-cesarean section abdominal wall endometriosis that were surgically treated at our institution. Seven of the nine patients had endometriosis tissue at the cesarean wound, and two had endometriosis tissue in the umbilical region and rectus abdominis muscle, unrelated to the wound.

Materials and Methods

This retrospective analysis included patients who had undergone cesarean section and developed abdominal

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Uncommon Endometriosis of C/S Wound

Case No.	Age at onset	Number of previous cesarean sections	Postoperative onset period (years)	Site of abdominal wall endometriosis	Depth of abdominal wall endometriosis	Complications of pelvic endometriosis	Clinical symptoms	CA125 (U/mL)
1	33	1	2	Wound	Subfascial	No	Pain	32
2	34	1	1	Wound	Subcutaneous	No	Pain	No record
3	35	1	1	Wound	Subfascial	No	Pain	30
4	39	1	1	Wound	Subcutaneous	Yes	Pain	49
5	40	1	2	Wound	Subcutaneous	No	Mass palpitation	No record
6	42	1	3	Wound	Subcutaneous	Yes	Pain/Mass palpitation	31
7	44	2	4	Outside of wound	Rectus abdominis muscle	No	Pain/Mass palpitation	34.1
8	45	3	4	Wound	Subcutaneous	No	Pain	35
9	45	1	18	Outside of wound	Whole umbilical region	Yes	Bleeding in the umbilical region	119.3

Table 1 Nine cases of abdominal wall endometriosis after cesarean section

wall endometriosis that required surgical treatment at our institutions during the period from April 2007 through August 2020. Nine patients who meet the criteria were identified in the medical records. Patients were included in the study if they had been diagnosed with abdominal wall endometriosis and had made an informed decision to undergo surgery after receiving an adequate explanation of hormone therapy, or if they had no improvement in symptoms with hormone therapy. Age at onset, number of cesarean sections, interval from cesarean section to onset of abdominal wall endometriosis, site of skin incision at cesarean section, site and depth of abdominal wall endometriosis, presence of pelvic endometriosis, presence of dysmenorrhea, other clinical symptoms, and cancer antigen-125 (CA-125) level were collected from medical records and retrospectively reviewed. The complication pelvic endometriosis was noted if a review of clinical findings and imaging studies, such as magnetic resonance imaging (MRI), confirmed that pelvic endometriosis was present at the time of the patient's outpatient visit for evaluation of wound endometriosis, rather than at the time of cesarean section. Written consent for case reporting was obtained from all patients.

Results

Table 1 shows the characteristics of the nine cases. The clinical diagnosis was based on clinical symptoms and ultrasound or MRI findings. No patient required cesarean section, and no patient showed evidence of endometriosis during a previous cesarean section. The mean age at onset of abdominal wall endometriosis was 39.7 years,

and all patients underwent 1-3 cesarean sections. The interval from the last cesarean section to endometriosis onset was approximately 1-4 years in most cases, but case 9 developed endometriosis 18 years after cesarean section. It was estimated that 6/9 (67%) of the cases had symptoms related to menstruation. Abdominal wall endometriosis was related to the cesarean procedure in 7/9 (78%) of cases. Abdominal wall endometriosis was located at the wound site in all five cases for which a longitudinal incision was used (Fig. 1). In contrast, endometriosis developed outside the skin wound in two cases and at the abdominal wall wound in two cases after a transverse incision (Fig. 1). The skin and fascia are commonly incised transversely, while the peritoneum is incised longitudinally in cesarean section (Pfannenstiel incision). The site of endometriosis in case 7 coincided with the most cephalic region of this longitudinal incision of the peritoneum of the transverse incision (Fig. 2 A). The depth of abdominal wall endometriosis was subcutaneous in five cases, subfascial in two cases, and to the rectus abdominis in one case. Endometriotic tissue was found in all layers of the umbilical region in case 9 (Fig. 2B). Typical pelvic endometriosis complications were noted in three cases; the other six patients had no endometriosis other than that in the abdominal wall. Five patients had dysmenorrhea, and three of these women had functional dysmenorrhea. The clinical symptoms that prompted the visit were lesion pain in seven cases and palpation of the mass in three cases. Bleeding from the lesion was the main complaint in the patient with umbilical endometriosis. CA125, a tumor marker for endometriosis, was preoperatively measured in seven pa-



(A) Five patients developed endometriosis lesions in the wound after cesarean section through a longitudinal incision, as shown by the red circles. (B) Four patients developed endometriosis lesions in the wound through a transverse incision after a cesarean section. Endometriosis occurred outside the cesarean wound in two cases, as shown by the blue circles.

tients; the mean value was 47.2 U/mL (reference value, $\leq 35 \text{ U/mL}$).

Diagnosis of abdominal wall endometriosis was based on clinical symptoms and imaging findings, and surgical resection was performed as treatment. The endometriosis lesion was excised with a margin of approximately 1 cm after marking the excision site (Fig. 3) as a joint operation with a plastic surgeon. In the case involving the rectus abdominis muscle, we designed a 2-cm circumferential margin, ligated the deep inferior abdominal wall artery, and then incised the rectus abdominis muscle to remove the lesion. The anterior sheath of the rectus abdominis muscle was sutured, after which the fascia and dermis were sutured. In one case, which occurred in the umbilical region, the lesion was excised with a margin of 1 cm. A fan-shaped skin valve was created in the umbilical region, and the upper and lower excess skin was excised. Both skin valves were sutured subcutaneously and the umbilicus was formed by inverting and suturing. In other cases, the lesions were resected with a 1-cm circumferential margin and sutured. No artificial repairs, such as synthetic mesh, were required in either case. All surgeries were performed without significant complications. Postoperatively, pathology specimens in all cases revealed endometriotic tissue consisting of endometrial glands and endometrial stromal cells (Fig. 4), which histologically confirmed abdominal wall endometriosis. No malignant findings were noted in any excised specimen.

Discussion

The site of endometriosis is classified pathologically as common, less common, and rare⁵. The wound, umbilicus, and groin are regarded as cutaneous tissue and classified as less common sites⁶. The incidence of endometriosis at a less common or rare site is 0.5% to several percent of all endometriosis cases in Japan⁷. The incidence of abdominal wall endometriosis is 0.03%-0.4% in patients undergoing cesarean section⁸.

A cesarean section requires a large endometrial incision through which the fetus, placenta, and amniotic membrane are removed. Therefore, in most cases, abdominal wall endometriosis after cesarean section is thought to result from implantation of endometrial tissue into the wound due to surgical manipulation. Additionally, the incidence of abdominal wall endometriosis after cesarean section before the onset of regular uterine contractions is approximately twice that after the onset of regular uterine contractions⁹. De Oliveria et al.¹⁰ reported that cesarean section before 22 weeks of gestation increased the risk of abdominal wall endometriosis. These findings suggest that immune tolerance is associated with subcutaneous adherent endometrial tissue that is not eliminated and grows back as endometriosis.

Methods of preventing abdominal wall endometriosis



- Fig. 2 A: MRI findings in patients with endometriosis outside the wound (case 7) Subtraction MRI image of case 7. A 4-cm high-contrast tumor is seen in the right rectus abdominal muscle on the cephalocaudal plane. A sagittal slice is shown on the left, and a horizontal slice is shown on the right.
 - B: MRI findings in patients with endometriosis outside the wound (case 9) T2-weighted MRI image of case 9. A 3-cm mass is seen in the umbilical region on the cephalocaudal plane. Additionally, multiple large myomas are present above the umbilicus, near the uterus. A sagittal slice is shown on the left, and a horizontal slice is shown on the right.

include using different sutures for the myometrium, peritoneum, fascia, and subcutaneous tissue during uterine surgery^{11,12}; thorough washing of the abdominal cavity and wound before closure of the abdomen¹³; avoiding the use of gauze or other materials that have been used to wipe the uterine lumen in the surgical field postoperatively¹¹, and protecting the uterus and preventing uterine tissue from being implanted¹². These intraoperative prophylactic measures are essential to prevent abdominal wall endometriosis, as the number of cesarean sections is increasing.

Only approximately 40% of patients with cutaneous endometriosis have symptoms associated with menstruation¹⁴. Endometriosis diagnosis and appropriate treatment might be delayed in patients without associated skin symptoms. The present patient 9 waited 3 years to be evaluated by an obstetrician/gynecologist, because she had no pain and no history of pelvic endometriosis. Pathologic confirmation by fine-needle aspiration biopsy is required if the patient does not present with typical symptoms and the diagnosis is difficult¹⁵.

Treatment of abdominal wall endometriosis includes surgery and endocrine therapy. Excision of the lesion may improve symptoms and allow pathologic diagnosis. Perioperative complications are minimal. The recurrence rate is relatively high, 4.5%-9.1%, when the extent of resection is inadequate⁴. A margin of approximately 1 cm is recommended for surgical resection, and disease recurrence after adequate resection is rare². However, reconstructive surgery with synthetic mesh or grafts may be necessary if the resection is large and the fascial defect is extensive. Close collaboration with a plastic surgeon is



Fig. 3 Resection marking for the endometriosis lesion and gross findings of resected specimens

(A) In case 7, subcutaneous tissue was exposed and the fascia was marked, after which the fascia and rectus abdominis muscle, where endometriosis was present, were excised en bloc. (B) Gross image of the excised specimen in case 7. (C) In case 9, the skin around the umbilicus was marked, and the entire abdominal wall, including the umbilicus, was excised. (D) Gross image of the excised specimen in case 9.

essential for surgical management of abdominal wall endometriosis. Conversely, conservative treatments, such as endocrine therapy, are useful². Hormonal therapy using progestin therapy or gonadotropin-releasing hormone analogs can help relieve symptoms and is used first when surgical treatment is not desired¹¹. However, hormone therapy alone is not definitive because symptoms often recur if treatment is discontinued¹⁶. If adequate resection margins cannot be achieved, postoperative hormone therapy may be considered to prevent recurrence.

Some studies have reported low efficacy for hormone therapy¹⁷ and development of malignancy¹⁸ in patients with abdominal wall endometriosis. Surgical excision of the lesion is a possible first-line treatment option, as it

has few perioperative complications, rapidly improves symptoms, and enables a pathological diagnosis. When symptoms are well-controlled with hormone therapy and there are no findings suggestive of malignancy, hormone therapy until menopause may also be considered.

Our study revealed that endometriosis at the cesarean wound was subcutaneous or subfascial in seven of nine cases. Among the two cases of endometriosis outside the wound, endometriosis in case 7 was likely caused by surgical manipulation of the uterine incision. The fascia and peritoneum are longitudinally incised after the transverse skin incision, and endometriosis can develop in the rectus abdominis muscle. In the present series, only case 9 is considered a rare-site endometriosis unrelated to surgery,



Fig. 4 Micrographs of excised endometriosis lesions (A) Histologically, ectopic endometrial glands with stromal tissue are seen. All 9 cases had similar findings, and abdominal wall endometriosis was diagnosed histologically. (B) Endometrial gland tissue showed some bleeding into the lumen.

as it was located in the umbilical region, away from the cesarean wound. Umbilical endometriosis, also known as Villar's nodule, was first reported in 188619. Although umbilical endometriosis is the most common form of cutaneous endometriosis²⁰, it accounts for only 0.4%-4% of extragenital endometriosis cases²¹. Pathologic evidence of endometrioid-like glandular ducts and stromal tissue in the umbilical region is required in order to confirm a diagnosis of umbilical endometriosis, as with other forms of endometriosis. When clinical symptoms such as umbilical pain and bleeding coincide with the menstrual cycle, a diagnosis of umbilical endometriosis may be made based on imaging studies such as CT or MRI and the effects of drug therapy alone. Hence, some cases are followed up as clinical umbilical endometriosis without pathologic examination. In the present cases 4 and 9, CA 125 was high. In case 4, the patient had endometriotic cysts in the ovaries and uterus, and in case 9, the patient had adenomyosis, so CA125 cannot be considered specific for abdominal wall endometriosis.

Surgical treatment of umbilical endometriosis often involves radical surgical therapy with local extended resection. Reconstruction in collaboration with plastic surgery is more common for the umbilical region than for other abdominal wall endometrioses.

Preventing abdominal wall endometriosis will become increasingly crucial as cesarean sections increase worldwide. Future developments will require risk stratification based on the number of weeks the cesarean section was performed and whether the operation was an emergency procedure. In addition, to limit invasiveness when surgical treatment will result in a sizable myometrial defect, regimens that combine hormonal therapy with surgery should be developed.

In conclusion, abdominal wall endometriosis may occur in women of reproductive age with a history of abdominal surgery, including cesarean section. Women with a history of abdominal surgery who present with recurrent pain or masses in the wound or abdominal wall that coincide with the menstrual cycle should be closely evaluated for abdominal wall endometriosis. Patients may be referred to dermatology or plastic surgery instead of obstetrics and gynecology if symptoms are nonspecific. Additionally, because surgical treatment often requires postoperative wound reconstruction, collaboration with plastic surgeons and dermatologists is essential.

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