Case Report of a Rare Case of Cesarean Scar Endometriosis

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Received Date: February 01, 2022
Published Date: March 03, 2022

Abstract

Scar endometriosis is a rare extra-pelvic form of endometriosis usually caused by iatrogenic seeding after obstetric or gynecological surgeries. Still, it can occur independently in any woman of childbearing age with a cyclic, painful lump in and around an obstetric or gynecologic scar after all other diagnoses have been ruled out. The preferred treatment is surgical excision, and a definitive diagnosis is made by histopathological assessment. We report a case of scar endometriosis in a young female who had two previous cesarean sections, and excision was planned along with the elective cesarean section, per patient request.

Keywords: CSE, endometriosis, Cesarean section scar.
Introduction

Endometriosis is an estrogen-dependent chronic disorder described first by Karl Von Rokitansky in 1860. (1) In this condition, the stromal tissue and functional endometrial glands are seen outside the uterine cavity, most commonly the pelvis, including the ovaries, the uterosacral ligaments, the pouch of Douglas, the bladder, distal ureter, and the rectosigmoid colon. Endometriosis can affect the heart, liver, kidney, lungs, central nervous system, and abdominal wall, in addition to the pelvic cavity. The most widely accepted theory of its formation is direct implantation. (2). Ectopic endometriosis foci are not usually malignant (2). Patients sometimes present to general surgery clinics with a painful lump. The diagnosis can be confused with abscess, lipoma, hematoma, sebaceous cyst, stitch granuloma, incisional hernia, or tumors when it appears in the abdominal wall, causing a delay in diagnosis. (3). The researchers determined that undergoing a cesarean section increased the risk of endometriosis. They found one extra case of endometriosis for every 325 women who had a cesarean section. However, it has been reported in episiotomy, hysterectomy, and laparotomy scars (4).

Case Report

A 37-year-old woman was seen in the outpatient Obstetrics and Gynecology department at our Arrayan Hospital on Jan 24, 2021. She presented for antenatal care. She was in her third pregnancy and had two children delivered by cesarean section. She had a history of asthma and an appendectomy. She had her first cesarean section in 2010 due to face presentation, and the second cesarean was done electively in October 2012. Her postoperative recovery was uneventful.

Next, she presented in Nov 2012 with an episode of pain left iliac fossa to the antenatal clinic. Her pregnancy test was negative, and urine analysis revealed mild urinary tract infection. She was given treatment for a urinary infection.

Later about five years after the cesarean section, in June 2017, she reported to a gastroenterologist with a history of intermittent lower abdominal pain. She had regular bowel habits, and there was no history of fever or weight loss. Examination revealed a small nodular mass of 1x2 cm size and firm in consistency, just beneath the scar at the left end. The cesarean scar was otherwise normal. The patient noted occasional regression of the mass. A provisional diagnosis of a query lipoma or hernia was made. An ultrasound abdomen was ordered, which showed a nonspecific hypoechoic mass of the subcutaneous fat with an impression of a dermatofibroma. (Figure 1).
The patient was referred to a general surgeon, who offered an excision, but she opted to wait. For two years, she had an intrauterine contraceptive device (IUCD) inserted for contraception.

On Nov 10, 2018, she presented with cyclical pain during her periods and wanted removal of IUCD. The device was removed, and she was offered excision, but she preferred to wait till the next delivery. Subsequently, she conceived spontaneously and followed in the antenatal clinic. Her pregnancy was unremarkable, and she was otherwise healthy. She had a history of two cesarean sections; therefore, she opted for excision at the elective cesarean section. Her expected delivery date was Aug 29, 2021, and a cesarean section (LSCS) was planned at 38 weeks gestation. Ultrasound was reviewed, and the case was discussed with the surgeon who decided excision on the same day as LSCS.

The patient was admitted on Aug 14, 2021, after preoperative checks, for the elective cesarean section. She delivered a healthy baby boy at 1117 hrs, with an Apgar of 9/10 and weighing 2650 grams. No intraabdominal pathology was found. (Figure 2).
The surgeon successfully performed the excision of the mass at the end of the cesarean section. Gross examination showed multiple tanned, yellow fibrofatty tissue fragments aggregating 4x 3x 2 cm. The specimen was sent to the laboratory for histopathological examination.

The patient was discharged from the hospital on the fourth postoperative day after a successful recovery. On follow-up in the clinic on the ninth postoperative day, she had been doing well, and her cesarean wound was clean and dry. Histopathology specimen revealed endometrial glands and stroma on fibro adipose and muscular tissue background. The report supported the diagnosis of scar endometriosis. (Figure 3). The patient has been advised for the regular follow-up to check for any recurrence.
Diagnosis:

"?? Scar tissue", excision:
- Endometriosis.

Comment:

The specimen consist of fibroadipose and muscular tissue. There are endometrial glands and stroma with extensive decidualization. The findings are consistent with endometriosis.

Gross Description:

The specimen is received in formalin labeled "?? Scar endometriosis" and consists of multiple tan-yellow fibrofatty tissue fragments aggregating 4.0 x 3.0 x 2.0 cm. The specimen is sectioned and representative sections are submitted in four cassettes.

Specimen Submitted:

?? Scar endometriosis.

Clinical History:

37-year-old female with swelling on side of previous scar.

Figure 3: Histopathology report confirming scar endometriosis.

Discussion

Scar endometriosis (CSE) is a rare condition with an incidence between 0.03% and 1.7%, and surgical scar endometriosis, which occurs after cesarean sections, has an incidence of 0.03-0.4%. (5).

A cyclical or non-cyclical painful mass is the most common finding. Endometrial or placental cells are transplanted into the wound during the surgical procedure. Hormonal changes influence this tissue. Estrogen stimulates it, resulting in discomfort, swelling, soreness, and discharge, all of which are common signs of scar endometriosis. (6).

Scar endometriosis is frequently misdiagnosed due to the wide range of signs and symptoms. This happened in our case, and as the patient got pregnant, her symptoms sometimes regressed.

Endometriosis is characterized by cyclical fluctuations in the size and intensity of discomfort during menstruation, but only 20% of patients suffer from these symptoms. (7). Our patient complained of pain related to her cycles in the pre-pregnancy period.

Even though endometrial cells are implanted at surgery, patients may present months or years later. The same happened in our case. Elabsi et al. described an abdominal wall endometrioma that developed 22 years after cesarean surgery. (8). In our report, the postsurgical period of the patients was eight years, and symptoms
appeared for the past four years. Even though scar endometriosis may occur three months to 10 years after gynecologic surgery, the mean occurrence period is 30 months.

Ultrasound with color Doppler, CT (Computed Tomography) scan, and MRI (Magnetic Resonance Imaging) can all be used to make the diagnosis, although these tests are not very specific. (9) Fine needle aspiration cytology is a fast, low-cost, and accurate diagnostic tool; however, there are some drawbacks, possibilities of seeding cells along the needle's tract. This could happen again in the future. (10) Ultrasoundography can provide information on the size, location, margins, and internal structure, even in a small mass. It can quickly distinguish between solid and cystic masses. A CT scan or an MRI can be employed if the diagnosis is in dispute. MRI can help define the anatomy of the soft tissue mass and its surrounding structures. Although the various imaging modalities are nonspecific, they help assess the degree of disease and aid in planning the surgical resection. Our patient had an ultrasound, but the diagnosis was not precise.

Medical therapy and surgery are the most common treatments for scar endometriosis. The main issue with medical management is the high recurrence rate after discontinuation, the side effects and interference with fertility. Surgical treatment provides the best opportunity for a definitive diagnosis and treatment of cesarean scar endometriosis. During excision, make sure at least clear margins of at least 1 cm away from the solid tissue should be included in the excision. (11) Furthermore, abdominal wall endometriosis incorporated into the abdominal wall musculature necessitates en bloc resection of the underlying myofascial elements. The excision site must be sterilized with neomycin or 0.9 percent sodium chloride solution. The abdominal wall can sometimes be rebuilt using a synthetic mesh to help to prevent a postoperative hernia. Histopathology is the most accurate method of diagnosing this condition.

The presence of endometrial-like glands spindled endometrial stroma, and hemosiderin accumulation inside macrophages or in the stroma all indicate endometriosis. (11).

On the other hand, preoperative medical treatment may be beneficial in reducing the lesion. Local recurrence is unpredictable and can occur, particularly after incomplete surgical excision. Bektas et al. discovered a 9.1 percent recurrence rate. Instead, when used as adjuvant hormonal therapy following surgical excision, it decreased recurrence from 42.9 percent to 11 percent. (12) Our patient was symptom-free following surgery; however, long-term follow-up will be required to confirm a complete cure.

Scar endometriosis is most common after previous cesarean sections, as in our case. Medical management was used in a case report; however, it was unsuccessful due to recurrence. Finally, surgical excision resulted in complete remission. (13) In another case report. (14), lesions were misdiagnosed as stitch granulomas. They were treated surgically, and histopathology confirmed the diagnosis.

Scar endometriosis is thought to develop due to iatrogenic auto-transplantation of endometrial cells during surgery. Apart from the pelvic organs, it can be found in the lungs, liver, kidneys, ureters, central nervous system, abdominal scar tissues, and extremities. The abdominal scar endometriosis has been noted after hysterectomy and excision of ovarian endometrioma. Endometriosis at the episiotomy site is also uncommon in
the literature. A case report described two rare cases of scar endometriosis: one after a laparotomy for a pregnancy in a rudimentary horn and the other in the scar from laparoscopic surgery for ovarian endometrioma (15).

One of the most accepted theories to describe CSE is the mechanical iatrogenic implantation theory. Endometrial cells inoculated directly into the surgical area can cause various clinical symptoms due to the proliferation of these cells under the influence of female hormones. Although CSE has many different clinical presentations, the most common symptoms and signs are cyclic pain, localized swelling, and tenderness. (16)

Risk factors of scar endometriosis have been studied and include hysterotomy at less than 22 weeks of pregnancy as a significant factor, heavy menstrual flow, and alcohol intake. (17) High parity has been considered a protecting factor. The main risk factor has been studied as obstetric surgery as it can expose many endometrial cells, which get entrapped in the wound. (18); especially noted in obese and overweight.

It has been postulated that failure to close the parietal and visceral peritoneum at the time of cesarean section may increase the risk of CSE. (19). Other strategies include not using a sponge to clean the endometrial cavity based on implantation theory. Strict cesarean delivery control is strongly advised. A high index of clinical suspicion is needed for early diagnosis and to avoid unnecessary referrals. Complete wide excision with clear margins can serve both diagnostic and therapeutic purposes.

**Conclusion**

CSE stands for cesarean scar endometriosis, a rare form of extra-pelvic endometriosis. Endometrial cells are implanted directly into the surgical region and, under ideal conditions, can proceed to endometriosis. When it is present in the abdominal wall, it can be confused with abscess, lipoma, hematoma, sebaceous cyst, stitch granuloma, incisional hernia, or tumors resulting in a delay in diagnosis. Although CSE can present in various ways in the clinic, the mainstay for early diagnosis is the awareness of the usual clinical symptoms. The diagnosis can be made on ultrasound with color Doppler, CT scan and MRI, but these investigations lack specificity. Patients who visit the hospital with symptoms of a mass on the abdominal wall, previous surgical history, and a painful mass becoming increasingly severe during menstruation periods must be questioned in full. Excision with a secure margin is necessary for the treatment of scar endometriosis. Special precautions should be taken during surgery to avoid spreading the endometrial tissue and covering the margins of the incision to form a physical barrier.

**Conflict of Interest:** No conflict of interest has been declared by any authors.

**Ethical permission is not** required. The case has been presented as anonymous.

**Author's contributions:** FA formal analysis and writing study conceptualization review and editing.
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Citation: Thana Bashir Goda. “Case Report of a Rare Case of Cesarean Scar Endometriosis.” MAR Gynecology 2.5 www.medicalandresearch.com (pg. 8)


Citation: Thana Bashir Goda. “Case Report of a Rare Case of Cesarean Scar Endometriosis.” MAR Gynecology 2.5
www.medicalandresearch.com (pg. 9)