Surgical Treatment of Scar Endometriosis Following Cesarean Section, a Series of 12 Cases

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Abstract It is difficult to conduct studies with larger series in rarely observed diseases. We report our experience in managing cesarean scar endometriosis (CSE) and emphasize the diagnosis and treatment options. The objective of our study is to review the clinical characteristics of CSE and to evaluate our surgical outcomes. We have collected and documented a case series of 12 patients who underwent surgical wide en bloc excision with surrounding clear margins for CSE. Patients' demographic features, symptoms, and clinical and operative findings were evaluated. The mean age was 34.6 years. Cyclical pain was documented in seven patients, while three patients presented with noncyclical pain. Menstrually-related enlargement of the nodule was observed in four patients, and only one patient had a complaint of dark brown leakage. The mean operation time was 26 min. The endometriotic lesions ranged from a diameter of 2 to 8 cm in size. Patients recovered completely, and no recurrence was observed. To prevent iatrogenic transplantation, additional attention is needed during surgery that exposes endometrial tissue. Complete wide excision of CSE is both diagnostic and therapeutic. To avoid unnecessary referrals, awareness of its typical clinical manifestations remains the mainstay for intervention. The most

important issues to be considered during surgery is nonspreading endometriosis while manipulation.

Keywords Endometriosis · Abdominal wall endometriosis · Cutaneous endometriosis · Scar endometriosis · Endometriosis surgery

Introduction

Endometriosis is a condition where the functional and morphological endometrial glands and stromal structures are found outside the uterus. It mainly affects women in reproductive ages. Although benign in structure, endometriosis has all the features of malignancy: local spread, invasiveness, and an outstanding ability to disseminate [1]. Endometriosis occurs most often in pelvis, on the surface lining of the pelvic cavity, peritoneum, ovaries, posterior cul-de-sac, and uterosacral ligaments. Rarely, implants of endometriosis can occur outside of the pelvis, and these forms are termed as extra pelvic endometriosis. It can also arise from scar tissues especially after cesarean section. The term cesarean scar endometriosis (CSE) is an unusual manifestation of extra pelvic endometriosis. One of the most accepted theory about CSE is the mechanical iatrogenic implantation. Endometrial cells are inoculated directly into the surgical area and can progress to endometriosis in optimal conditions. This causes various clinical symptoms due to proliferation of these cells under the influence of female hormones. Although CSE has many different clinic presentations, the most common clinical symptoms and signs are swelling, tenderness on local site, and cyclic pain. Wide en bloc excision with surrounding clear margins is both diagnostic and therapeutic intervention.

The aim of this article is to review the clinical characteristics of CSE and to assess our surgical outcomes. The diagnosis

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and treatment options were analyzed, and the literature regarding this entity was discussed.

Methods

We have collected and documented a case series of 12 patients who underwent surgical management for CSE in our obstetrics and gynecology clinic at Ümraniye Education and Research Hospital between January 2006 and December 2011. All patients were informed about surgical management and written informed consents were obtained. All patients had a history of previous cesarean section, and their initial cesarean sections were performed in different hospitals. After the clinical assessment, the diagnosis was confirmed by pelvic ultrasonography.

We performed sharp dissection with a scalpel within the area of the incision from the previous cesarean section (Fig. 1). The cystic mass which was surrounded by fibrosis was removed carefully with a safe margin (Figs. 2 and 3). The surgical intervention was followed by the reconstruction of the abdominal wall in anatomical layers.

All in cases, surgical wide en bloc excision with surrounding clear margins was performed (Figs. 2 and 3), and the definitive diagnosis was confirmed by the pathological examination. All patients were operated under spinal anesthesia. Age, parity, body mass index, symptoms, size of tumor, time between cesarean section and the onset of symptoms, operative findings, and surgical outcomes were evaluated. Demographic features and operative findings of the cases are demonstrated in Table 1.

We state that the protocol for the research project has been approved by a suitably constituted Ethics Committee of the institution within which the work was undertaken and that it conforms to the provisions of the Declaration of Helsinki.

Fig. 1 The initial incision within Pfannenstiel incision

Fig. 2 The excised tumoral mass

Results

This study includes the medical records of 12 patients who underwent surgical treatment for CSE. The first admissions of seven patients were to gynecologists, four to general surgeons, and one to a dermatologist. All were referred to our territory health care institution. Based on medical records, Pfannenstiel incision had been performed for cesarean section in all patients. The median age was 34.6 ± 5.43 years (range from 24 to 44 years), and the mean BMI was 26.95 ± 3.59 kg/m² (range from 22.30 to 33.50 kg/m²). The common complaint of the patients was a palpable subcutaneous mass under the incision scar. Seven patients suffered from cyclical pain. Noncyclic pain was seen in three patients, and four patients had experienced the enlargement of the nodule during the menstrual period. Only one patient had a complaint of menstrually-related dark brown leakage. The mean time interval between initial cesarean section and the onset of symptoms was 45 months (range of 16 to

Fig. 3 Gross appearance of a cut-open specimen

Table 1 Demographic features and operative findings of cases

Cases	Age (years)	Gravida	Parity	Body mass index (BMI; kg/m ²)	Previous abdominal surgeries	Asymptomatic period (months)	Operation time (min)
1	33	2	2	29.8	Cesarean section (2)	36	21
2	37	3	4	23.1	Cesarean section (2) appendectomy	72	28
3	35	3	2	22.3	Cesarean section (2)	108	23
4	24	1	1	30.1	Cesarean section	16	16
5	30	2	1	29.7	Cesarean section salpingectomy	24	26
6	44	3	3	24.1	Cesarean section (3)	42	32
7	34	2	2	23	Cesarean section (2)	48	22
8	39	2	1	24.7	Cesarean section	42	24
9	36	3	3	33.5	Cesarean section (3)	30	29
10	34	4	3	26.9	Cesarean section (3)	54	46
11	29	3	2	28.1	Cesarean section (2)	36	18
12	41	2	2	29.8	Cesarean section (1)	30	27

Asymptomatic period was defined as the time interval between the previous surgery and the onset of the symptoms. Number in the parentheses indicates the number of procedures

108 months). The preoperative diagnosis was correct in all patients. Two patients had medical treatment before admission to our clinic. Those two patients had a history of medical treatment failure. All of the patients were treated surgically. Almost all the nodules were excised easily. In five cases, extensions of the lesions through the facial layer were seen during surgery. In these cases, facial defects were repaired after excision (Fig. 4). The mean operation time was 26.26 ± 7.79 (16–46) min. The diameter of the endometriotic lesions ranged from 2 to 8 cm in size. All patients were completely recovered without relapse of symptoms. The follow-up period ranged from 12 to 60 months. No recurrence was observed during the follow-up period. Indications for cesarean section were previous cesarean section (5), fetal distress (2), cephalopelvic disproportion (2), breech presentation (1), twin pregnancy (1), and eclampsia (1). The mean number of parity was 2.1 with the range of 1 to 4.

Discussion

Endometriosis in cesarean scar is a rarely observed disease. There are a limited number of publications focusing on CSE,

Fig. 4 The view of repaired facial defects

and most of them are case reports. It is difficult to perform well-controlled clinical trials in rarely observed disease. The common presentation of CSE includes palpable subcutaneous mass, typically accompanied by cyclic, noncyclic, or constant pain. Menstruation usually aggravates disease. The mass under a cesarean section scar and the symptoms in a cyclic manner fairly facilitate the diagnosis of the illness.

De Oliveira et al. reported a case-control study to identify the risk factors of scar endometriosis. According to this study, early hysterotomy in pregnancy especially before 22nd week of gestation is the main risk factor [2]. Additionally, increased menstrual flow and alcohol consumption are also concluded as risk factors, while high parity may be a protecting factor [2]. The most evident risk factor for the presence of endometriosis in scar tissue is a previous history of obstetric surgical procedures [3]. Why obstetric surgery may be an important risk factor in contributing to wound endometriosis? The possible causes were defined by Wang et al. First of all, obstetric surgery can expose a large amount of endometrial cells, and these cells can be entrapped in the wound [4]. The separation of active cells may be facilitated by amniotic fluid and significantly more blood loss in obstetric surgery would provide a relatively rich nutritional environment for the growth of endometrial tissue in the wound [4]. In this study, more than half of our patients were obese and overweight. Obesity can provide wide surgical surface for the entrapment of endometrial active cells and may be an impact on the illness.

In this study, the time interval between cesarean section and the onset of symptoms is ranged from 16 months to 9 years. The late onset of symptoms after surgery is the probable cause of misdiagnosis [1]. The mean duration of the symptoms was higher in patients whom first admission was not to the obstetrician. Overall, general surgeons are infrequently involved in

the management of cesarean section scar lesions [5]. Preoperative diagnoses of surgical scar endometriosis are often wrong, especially those made by general surgeons [3]. Physical examination is essential for an accurate diagnosis. Scar endometriosis is usually developed in superficial layers of the connective tissue, and nodules are usually found by palpation. The clinical evaluation can be confirmed by pelvic ultrasonography. Ultrasound is the most accessible, reliable, and cost-effective procedure. Some additional diagnostic procedures such as fine-needle aspiration cytodiagnosis, computed tomography, and magnetic resonance imaging can be performed [5, 6]. The imaging modalities are nonspecific and more useful for differential diagnoses and detecting the relationship between the mass and the other tissues. These are also used in planning of operative resection, to identify and to evaluate the extent of disease. Awareness of its typical clinical manifestations remains the mainstay for intervention and diagnosis.

In this study, all cesarean sections were performed outside of our clinic. Hence, we could not contribute to the literature about the incidence of CSE. It can be assumed as a methodological limitation of our study. Based on literature reviewed, the estimated incidence of scar endometrioma ranges from 0.03 to 3.5 % and more frequently observed with an incidence of 0.03 to 0.47 % following cesarean delivery [3, 7]. Nominato et al. suggested that cesarean section greatly increases the risk of developing scar endometriosis [8]. It should be suspected in any woman, with scar-related masses, who had a history of uterine surgery, especially cesarean section. However, endometriosis on the abdominal wall can be observed in some sporadic cases that have not been previously exposed to any type of surgery [9]. Furthermore, some presentations of abdominal wall endometriomas are referred to surgery performed by general surgeons such as appendectomy [10]. In these cases, hematogenous outspread, lymphatic dissemination, coelomic metaplasia, and some else theories could be addressed for probable pathogenesis. In our study, all cases had a history of at least one cesarean section. By the fact that the number of cesarean sections is constantly increasing, this complication becomes more frequent [11].

In this case series, we did not find any evidence of pelvic endometriosis. Iatrogenic mechanical transplantations on incision scars during the operations are the most accepted pathogenesis. Aggressive behavior of endometriosis itself might also be a possible risk factor [4]. Mostly, the reported cases have mentioned that the contamination have occurred during surgery that possible contact with endometrial tissue including episiotomy, hysterectomy, ectopic pregnancy, laparoscopy, tubal ligation, and cesarean section [12, 13]. So it is important to take some precautions to avoid transplantation of endometrial cells. To minimize endometriosis contamination, some authors recommend careful isolation of the wall incision and lavage with saline before the closure of the wall [14]. The

others hypothesized that failure to close the parietal and visceral peritoneum with sutures at the time of cesarean section may markedly increase the postoperative occurrence of an endometrioma in the skin incision scar [15]. Replacing instruments and needles with a new one is recommended when suturing other abdominal layers [16]. Based on literature reviewed, we did not find any correlation between the indications of cesarean section and CSE. On the other hand, some authors reported the correlations with timing of cesarean section and CSE. Wicherek et al. stated that cesarean section performed before spontaneous onset of labor may increase substantially the risk of occurrence of scar endometriomas [17]. Immune tolerance during pregnancy was suggested to be an important factor predisposing to the implantation [17]. Similarly in this study, 8 of 12 patients had cesarean section without the presence of regular uterine contractions. In other words, eight cases were elective.

Medical treatment gives only partial relief and with regard to the almost certain recurrence of the condition after cessation of medication [5, 18]. The use of progestogens, oral contraceptive pills, and danazol are not effective [5]. Due to side effects of androgens, patients have poor compliance to these drugs [5]. Two of our patients had a history of medical treatment failure before admission to the clinic. Treatment of choice is wide excision. Surgical treatment is overly recommended [1, 19, 20].

The mean operation time was 26 min. None of them have required further surgical intervention in the follow-up. So in order to prevent the recurrences, surgical excision remains the preferable method. Local recurrence is likely to be after an inadequate surgical excision [1, 20].

As a conclusion, we noticed that cesarean section is an apparent risk factor for the presence of endometriosis. To prevent iatrogenic transplantation, additional attention is needed during surgery that exposes endometrial tissue. To avoid unnecessary referrals, awareness of its typical clinical manifestations remains the mainstay for intervention. Complete wide excision with clear margins is both diagnostic and therapeutic. The operation can be easily applied in a short time as well. One of the most important issue to be considered during surgery is nonspreading endometriosis while manipulation.

Conflict of Interest We declare that we have no conflicts of interest and no funding was received for this work.

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