Cesarean scar endometriosis: presentation of 198 cases and literature review

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Abstract

Background: Cesarean scar endometriosis (CSE) is the most common type of abdominal wall endometriosis (AWE). The aim of this study was to systematically identify the clinical features of CSE and recommend precautionary measures.

Methods: A large, retrospective study was undertaken with CSE patients treated surgically at our hospital between January 2005 and December 2017.

Results: A total of 198 CSE patients were enrolled, with a mean age of 32.0 ± 4.0 years. The main complaint of the patients was abdominal mass (98.5%), followed by cyclic pain (86.9%). The latency period of CSE was 31.6 ± 23.9 months, and the duration between the onset of symptoms and this surgery was 28.3 ± 25.0 months. A majority (80.8%, n = 160) of the patients had undergone a Pfannenstiel incision, and a minority (19.2%, n = 38) a vertical midline incision. The latency period of CSE in the case of a Pfannenstiel incision was significantly shorter than that in the case of a vertical midline incision (24.0 vs 33.0 months, P = 0.006). A total of 187 (94.4%) patients had a single endometrioma, 11 (5.6%) patients had multiple endometriomas, and the 11 multiple-endometrioma patients had all undergone a Pfannenstiel incision. Lesions of endometrioma were common in corner sites, after either incision: 142/171 (83.0%) in Pfannenstiel incision scars and 32/38 (84.2%) in vertical incision scars.

Conclusions: The findings of this study indicate that the Pfannenstiel incision carries a higher risk of CSE than the vertical midline incision. Thorough cleaning at the conclusion of CS, particularly of both corner sites of the adipose layer and the fascia layer, is strongly recommended for CSE prevention. Further studies might provide additional recommendations.

Keywords: Cesarean scar endometriosis, Abdominal wall endometriosis, Cesarean section, Pfannenstiel incision

Background

Endometriosis is a sex hormone-dependent gynecological disease that is characterized by the growth of endometrial tissue outside the uterine cavity [1]. It usually occurs in the pelvis, at sites such as the ovaries and the pelvic peritoneum. However, ectopic endometrial tissue can also be found outside the pelvis, at sites such as the lung, brain, bowel, and abdominal wall [2–4]. The presence of ectopic endometrial tissue embedded in the subcutaneous adipose layer and the muscles of the abdominal wall is called abdominal wall endometriosis (AWE). AWE can occur spontaneously, but usually develops in association with a previous surgical procedure, such as a cesarean section (CS), hysterectomy, or appendectomy [5–7].

Cesarean scar endometriosis (CSE) is the most commonly reported type of AWE [8]. Nominato et al. suggested that CS greatly increased the risk of developing AWE [9]. The pathophysiology of CSE may be due to the direct implantation of endometrial tissue in the cesarean incision (the implantation theory) [8]. During cesarean delivery, the endometrial tissue is inoculated directly in the cesarean incision. With an appropriate supply of nutrients and hormonal stimuli, these endometrial cells survive and proliferate, which finally leads to CSE. Although it is an unusual disease, with a reported incidence of 0.03–0.45%, CSE may cause long-term discomfort involving cyclic lower abdominal pain [10, 11]. Case reports of malignant transformation of CSE have also been sporadically reported [12–14].
Generally, few publications have focused on CSE, and a majority of them are either case series or case reports [15–18]. As a large obstetric and gynecologic hospital, we have treated many CSE patients and were very interested in identifying the characteristics associated with CSE. Here, we report our findings in managing CSE over approximately the last decade, and we discuss the clinical features and prevention strategies of this rare disease.

Methods
This retrospective study was conducted in the Department of Obstetrics and Gynecology of the International Peace Maternity and Child Health Hospital (IPMCH). The study was approved by the ethical review committee of the IPMCH.

This study included patients diagnosed with CSE who received surgical treatment at IPMCH between January 2005 and December 2017. The inclusion criteria were the following: (1) patient had a history of at least one CS and the symptoms occurred after the cesarean delivery; (2) surgical excision with surrounding clear margins was performed; and (3) pathological diagnosis for each excised lesion was endometriosis.

The baseline characteristics and surgical processes of all of the patients were recorded in the database of our hospital. We conducted a computerized search of the database according to the disease name. The preliminarily selected items were further checked and screened artificially following the inclusion criteria. The main symptoms of CSE included a palpable mass, under or away from the scar, with cyclic pain and swelling during menstruation. The latency period was defined as the time between CS and the onset of symptoms. The following information was extracted: age, age at CS, parity, delivery history, incision type, symptoms, size of the mass, latency period, duration between symptoms and surgery, operative findings, and histopathological evaluations.

The normality of continuous variables was tested using the Shapiro–Wilk test. The comparisons of normally distributed, continuous data were made with the Student t test and an analysis of variance. Two sets of nonnormally distributed, continuous data were analyzed with Mann–Whitney U tests; otherwise, Kruskal–Wallis H tests were employed. The categorical data were analyzed with \( \chi^2 \) and Fisher exact tests. The differences were considered statistically significant when the P-value was less than 0.05. Statistical analysis was performed using SPSS version 17.0 (SPSS Inc., Chicago, IL, USA).

Results
Following the inclusion standards described above, a total of 198 CSE cases were ultimately enrolled and all the cases were pathologically confirmed. Baseline characteristics of the patients are summarized in Table 1.

The mean age of the patients was 32.0 ± 4.0 years, and the mean age at the time of CS was 27.1 ± 3.5 years. Parity of the patients ranged from 1 to 2. All of the patients had a history of CS: 186 (93.9%) patients had 1 CS, and 12 (6.1%) had 2. The latency period of CSE ranged from 1 to 120 months, with a mean of 31.6 ± 23.9 months. The duration between the onset of symptoms and surgery was 28.3 ± 25.0 months.

Symptoms and CSE sites of the study patients are also shown in Table 1. The most common symptom of the patients was a palpable painful abdominal mass under or away from the scar, with cyclic pain and swelling during menstruation. The latency period was defined as the time between CS and the onset of symptoms.
adjacent to the incision, accompanied with either cyclic pain (86.9%, \( n = 172 \)) or noncyclic pain (13.1%, \( n = 26 \)). A majority (80.8%, \( n = 160 \)) of the patients had undergone a Pfannenstiel incision, and a minority (19.2%, \( n = 38 \)) a vertical midline incision. In the group of 198 study patients, 187 (94.4%) patients had a single endometrioma and 11 (5.6%) patients had multiple endometriomas. The 11 multiple-endometrioma patients had all undergone a Pfannenstiel incision. In total, 209 abdominal wall endometriomas were excised. Specifically, 171 and 38 endometriomas were excised from Pfannenstiel incision and vertical midline incision scars, respectively. A majority of the endometriomas were located in corner sites, after either incision: 142/171 (83.0%) in Pfannenstiel incision scars and 32/38 (84.2%) in vertical incision scars.

We introduced an “upper bound” and a “lower bound” to describe the locations of the endometriomas for the first time. The abdominal wall was divided into the adipose layer, the fascia layer, the muscular layer, and the peritoneal layer. The bladder was considered the deepest layer involved. As shown in Tables 2, 64.6% (\( n = 135 \)) of the endometriomas were located between the adipose layer and the fascia layer; 14.8% (\( n = 31 \)) were located between the adipose layer and the muscular layer. Of the 209 endometriomas, 16 (7.7%) invaded the peritoneum; 1 (0.5%) invaded into the abdominal cavity; and 2 (1.0%) invaded the bladder.

| Location           | AL, n (%) | FL, n (%) | ML, n (%) | PL, n (%) | AC, n (%) | B, n (%) |
|--------------------|-----------|-----------|-----------|-----------|-----------|---------|
| Upper bound        | AL        | FL        | ML        | PL        | AC        | B       |
| AL                 | 12 (5.7)  | 135 (64.6)| 31 (14.8) | 11 (5.3)  | 1 (0.5)   | 1 (0.5) |
| FL                 | /         | 0 (0)     | 12 (5.7)  | 3 (1.4)   | 0 (0)     | 0 (0)   |
| ML                 | /         | /         | 0 (0)     | 2 (1.0)   | 0 (0)     | 1 (0.5) |

**Table 2** Location of the endometriomas in the abdominal wall

\( AL = \) adipose layer, \( FL = \) fascia layer, \( ML = \) muscular layer, \( P = \) peritoneum layer, \( AC = \) abdominal cavity, \( B = \) bladder

Discussion

CSE is an uncommon iatrogenic disease caused by endometrium implantation in the incision during cesarean delivery. In the present study, we investigate 198 cases of CSE over a period of 13 years, providing detailed information that helps us to better understand the clinical characteristics of this rare condition.

Several theories about the pathogenesis of AWE have been proposed, such as the implantation theory, the coelomic metaplasia theory, and the lymphatic or hematogenous dissemination theory [8, 19]. As the most common type of AWE, CSE is best explained by the iatrogenic direct implantation theory. During cesarean delivery, endometrial tissue is seeded into the wound. With an appropriate supply of nutrients and hormonal stimuli, these endometrial cells survive and proliferate, finally leading to CSE. In the present study, most of the endometriomas were located in a corner of the incision scar: 83.0% in Pfannenstiel incision scars and 84.2% in vertical midline incision scars. In another large retrospective study, conducted by Yan Ding et al., similar results were obtained [20]. In their study, 77.1% of the endometriomas were located in the corners of the scars. This is probably because endometrial cells are less easily removed from the corners of the incisions during CS.

To identify the risk factors for CSE, we calculated the difference in latency period based on the patients’ baseline characteristics (Table 3). Age at CS, parity, previous CS, and dysmenorrhea showed no significant correlation with the latency period. Location of the endometriomas, such as under or away from the scar, or in the corner or in the middle of the scar, also showed no correlation with the latency period. However, the latency period showed significant correlation with the incision type, i.e., the latency period of the CSE in patients with Pfannenstiel incision was significantly shorter than that in patients with vertical midline incision (24.0 vs 33.0 months, \( P = 0.006 \)). The location of the lower bound of the endometriomas also showed a correlation with the latency period (\( P = 0.011 \)), i.e., the latency period was longer when the endometriomas invaded down to the peritoneum or the bladder.

To confirm the difference in latency period based on incision type, baseline characteristics of the patients with Pfannenstiel incisions or vertical midline incisions were further compared. As shown in Table 4, no significant difference was identified in the patients’ baseline characteristics.
incisional hernia and results in a better cosmetic appeal. However, the Pfannenstiel incision usually involves more dissections, and the blood loss following dissection may be greater [22].

CSE is a complication of cesarean surgery. Unfortunately, the relationship between the CS incision type and the pathogenesis of CSE is still unknown. The Pfannenstiel incision is the most commonly reported type for the occurrence of CSE; however, because of the disease rarity and the need for the pathological confirmation of the diagnosis, it is difficult to estimate the population-wide incidence of CSE for different incision types [19, 20]. Demiral et al. speculated that Pfannenstiel incisions confer a higher risk of CSE than do midline incisions, but

| Characteristic                        | n (%) | Latency period (months) | P-value |
|---------------------------------------|-------|-------------------------|---------|
|                                       |       | Median (quartiles)      |         |
| Age at CS (years)                     |       |                         |         |
| ≥ 35                                  | 6 (3.0)| 30.0 (10.5–48.0)        | 0.941   |
| 25–34                                 | 143 (72.2)| 24.0 (12.0–40.0)       |         |
| ≤ 24                                  | 49 (24.8)| 24.0 (12.0–48.0)        |         |
| Parity                                |       |                         | 0.273   |
| Nulliparous                           | 178 (89.2)| 24.0 (12.0–36.0)       |         |
| Multiparous                           | 20 (10.1)| 21.0 (6.0–48.0)         |         |
| One previous CS                       |       |                         | 0.452   |
| Yes                                   | 21 (10.6)| 24.0 (60–48.0)          |         |
| No                                    | 177 (89.4)| 24.0 (12.0–36.0)       |         |
| Dysmenorrhea                          |       |                         | 0.473   |
| Yes                                   | 64 (32.3)| 19.0 (12.0–36.0)       |         |
| No                                    | 134 (67.7)| 24.0 (12.0–36.0)       |         |
| Incision type                         |       |                         | 0.006   |
| Pfannenstiel                          | 160 (80.8)| 24.0 (12.0–36.0)       |         |
| Vertical midline                      | 38 (19.2)| 33.0 (24.0–60.0)        |         |
| No. of endometriomas                  |       |                         | 0.078   |
| Single                                | 187 (94.4)| 24.0 (12.0–48.0)       |         |
| Multiple                              | 11 (5.6) | 16.0 (12.0–24.0)        |         |
| Location of the endometriomas I      |       |                         | 0.253   |
| Under the scar                        | 111 (56.1)| 24.0 (12.0–36.0)       |         |
| Away from the scar                    | 87 (43.9)| 24.0 (12.0–48.0)        |         |
| Location of the endometriomas II     |       |                         | 0.153   |
| Corner of the scar                    | 167 (84.3)| 24.0 (12.0–48.0)       |         |
| Middle of the scar                    | 31 (15.7)| 30.0 (24.0–38.0)        |         |
| Upper bound of the endometriomas     |       |                         | 0.073   |
| Adipose layer                         | 181 (91.4)| 24.0 (12.0–39.0)       |         |
| Fascia layer                          | 14 (7.1) | 24.0 (18.0–49.5)        |         |
| Muscular layer                        | 3 (1.5) | 48.0 (48.0–56.0)        |         |
| Lower bound of the endometriomas     |       |                         | 0.011   |
| Adipose layer                         | 10 (5.1) | 27.0 (18.0–39.0)        |         |
| Fascia layer                          | 130 (65.7)| 24.0 (12.0–36.0)       |         |
| Muscular layer                        | 41 (20.7)| 24.0 (16.0–48.0)        |         |
| Peritoneum                            | 12 (6.1) | 48.0 (27.0–60.0)        |         |
| Abdominal cavity                      | 3 (1.5) | 30.0 (90–30.0)          |         |
| Bladder                               | 2 (1.0) | 40.0 (24.0–40.0)        |         |

*Mann–Whitney U test for comparing two sets; Kruskal–Wallis H test for comparing multiple sets*
An incision. Another interesting result from this study is that
ably carries a higher risk of CSE than the vertical midline
findings demonstrate that the Pfannenstiel incision prob-
endometriomas had Pfannenstiel incisions. These research
explanation, all 11 patients in this study who had multiple
cells, favoring the occurrence of CSE. Consistent with this
for the implantation and growth of residual endometrial
would provide a relatively rich nutritional environment
important role in the pathogenesis of endometriosis [25].
Therefore, more blood loss in the Pfannenstiel incision
section, more capillaries are cut off during a Pfannenstiel
longitudinal pattern of the abdominal vessels and the large dis-
second cause is a larger nutrient supply. Due to the longi-
dicular incisions. This indicates that, compared to the vertical
incision, the Pfannenstiel incision might be more favorable
to the implantation and proliferation of the residual endometrial cells. We suggest two possible causes for the
favorable role of the Pfannenstiel incision. First, the Pfan-
steniel incision involves wider dissection planes and more gaps, rendering tissue irrigation difficult and indu-
ing much more endometrial cell contamination [22]. The second cause is a larger nutrient supply. Due to the longi-
tudinal pattern of the abdominal vessels and the large dis-
section, more capillaries are cut off during a Pfannenstiel
incision than in a vertical incision, causing more blood
loss. Endometrial cells require an adequate blood supply
to survive in their ectopic sites, and angiogenesis plays an
important role in the pathogenesis of endometriosis [25].
Therefore, more blood loss in the Pfannenstiel incision
would provide a relatively rich nutritional environment
for the implantation and growth of residual endometrial
cells, favoring the occurrence of CSE. Consistent with this
explanation, all 11 patients in this study who had multiple
endometriomas had Pfannenstiel incisions. These research
findings demonstrate that the Pfannenstiel incision prob-
ably carries a higher risk of CSE than the vertical midline
incision. Another interesting result from this study is that
deeper endometrioma locations are correlated with longer
latency periods. This is probably due to the fact that the
deeper endometriomas could not be easily noticed.
Although CSE is a rare event, it manifests as a painful
subcutaneous mass and usually bothers the patient for several years. Additionally, CSE can undergo malignant
change, which is rapidly fatal and has a survival rate of
only 57% [14]. Hence, it is necessary to take precautions
to prevent or reduce the occurrence of CSE. On the
basis of the implantation theory, we propose a variety of
measures: careful flushing and irrigating before closure;
using separate needles for uterine and abdominal closure;
and not using a sponge to clean the endometrial cavity following complete delivery. Extending the breast-
feeding period to delay menstruation has also been pro-
posed for preventing CSE, but without scientific corroboration [21]. In our study, 83.3% (174/209) of the
scar endometriomas were located in corner sites of the
wound. Therefore, the abdominal wound should be
cleaned thoroughly with saline solution before closure,
especially the corner sites. Additionally, endometriomas
were more common in superficial parts of the abdominal
wall, i.e., 12/209 (5.7%) were present in the adipose layer
and 135/209 (64.6%) were present in the adipose layer and
the fascia layer, accounting for 70.3% of the total endome-
triomas. Therefore, careful flushing and irrigation of the
adipose layer and fascia layer during closure is critical.
All of the patients in our study underwent surgical ex-
cision for the treatment of CSE. Generally, surgical treat-
ment offers the best chance for both making a definitive
diagnosis and treating CSE. Medical therapy has a low
success rate is associated with adverse effects.
As a retrospective study, some limitations in this study
could not be avoided. For example, the data about the
CS procedures lacked details such as the layers of clos-
ure, type of suture materials, and operation duration.
These factors might also affect the occurrence of CSE.
To address these questions, further studies will be re-
quired in the future.

**Conclusions**

Concerning the rising CS rate, CSE may occur more fre-
quently than generally assumed. Early diagnosis, treat-
ment, and prevention of CSE are worthy of our
attention. In our large, retrospective study, we systema-
tically reviewed the clinical features of CSE and we pro-
vide the first evidence that the Pfannenstiel incision
carries a higher risk of CSE than the vertical midline
incision. The findings in this study will help us to better
understand CSE and devise precautionary measures to
reduce the occurrence of the disease.

**Abbreviations**

AWE: Abdominal wall endometriosis; CS: Cesarean section; CSE: Cesarean scar endometriosis

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**Table 4** Comparison of the baseline characteristics between patients with Pfannenstiel incision or vertical midline incision

| Characteristic          | Pfannenstiel | Vertical midline | P-value |
|-------------------------|--------------|------------------|---------|
| Age at CS (years)       |              |                  | 0.334   |
| ≥ 35                    | 4 (2.5)      | 2 (5.2)          |         |
| 25–34                   | 119 (74.4)   | 24 (63.2)        |         |
| ≤ 24                    | 37 (23.1)    | 12 (31.6)        |         |
| Parity                  |              |                  | 0.195   |
| Nulliparous             | 146 (73.7)   | 32 (16.2)        |         |
| Multiparous             | 14 (7.1)     | 6 (3.0)          |         |
| One previous CS         |              |                  | 0.248   |
| Yes                     | 15 (7.6)     | 6 (3.0)          |         |
| No                      | 145 (73.2)   | 32 (16.2)        |         |
| Dysmenorrhea            |              |                  | 0.782   |
| Yes                     | 51 (25.8)    | 13 (6.6)         |         |
| No                      | 109 (55.1)   | 25 (12.6)        |         |

*Fisher exact test* without sufficient evidence [23]. In this study, the latency period of CSE was 31.6 ± 23.9 months, which was comparable with that reported in other studies [19, 24]. However, when comparing the latency period of CSE in patients with Pfannenstiel incisions to those with vertical midline incisions, we observed a significantly shorter latency in Pfannenstiel incisions (24.0 vs 33.0 months, *P* = 0.006) (Table 3). In other words, CSE in patients with Pfannenstiel incisions occurred earlier than in patients with vertical incisions. This indicates that, compared to the vertical incision, the Pfannenstiel incision might be more favorable to the implantation and proliferation of the residual endometrial cells. We suggest two possible causes for the favorable role of the Pfannenstiel incision. First, the Pfannenstiel incision involves wider dissection planes and more gaps, rendering tissue irrigation difficult and indu-
ing much more endometrial cell contamination [22]. The second cause is a larger nutrient supply. Due to the longi-
tudinal pattern of the abdominal vessels and the large dis-
section, more capillaries are cut off during a Pfannenstiel
incision than in a vertical incision, causing more blood
loss. Endometrial cells require an adequate blood supply
to survive in their ectopic sites, and angiogenesis plays an
important role in the pathogenesis of endometriosis [25].
Therefore, more blood loss in the Pfannenstiel incision
would provide a relatively rich nutritional environment
for the implantation and growth of residual endometrial
cells, favoring the occurrence of CSE. Consistent with this
explanation, all 11 patients in this study who had multiple
endometriomas had Pfannenstiel incisions. These research
findings demonstrate that the Pfannenstiel incision prob-
ably carries a higher risk of CSE than the vertical midline
incision. Another interesting result from this study is that
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Availability of data and materials
The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors’ contributions
PZ proposed the study concept and design, assisted by HX, YBS, YPY, and LNZ and acquired and checked the data. CZ and NLW carried out the data analyses. PZ drafted the manuscript. All of the authors approved the final version of the manuscript.

Ethics approval and consent to participate
The study was approved by the ethical review committee of the International Peace Maternity and Child Health Hospital (IPMCH).

Consent for publication
Consent was obtained from the patients for the publication of this report.

Competing interests
The author declares that they have no competing interests.

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