

Abdominal wall endometriosis occurring after cesarean section: an underestimated complication

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Summary

The aim of the study was to review patients characteristics, describe the exact anatomic locations and size of the endometriosis in the abdominal wall, and discuss the factors that may contribute to mesh use during abdominal wall endometriosis (AWE) resection. *Materials and Methods:* Patients diagnosed with AWE in their surgical scars from January 2008 to December 2014 were documented. Descriptive data was collected and analyzed. *Results:* A total of 95 patients with an age ranging from 26 to 48 years, with a mean age of 33.5 ± 5.0 years at the time of excision were analyzed. The mean diameter of the mass was 3.25 cm in the present series with an average of 4.97 cm in the mesh group by ultrasound. A total of 18 patients had mesh therapy for fascia defect compared with 77 non-mesh therapy patients. The size of the lesions, the mean duration of symptoms for painful mass, and level of the serum CA125 were statistically different between mesh group and non-mesh group ($p < 0.05$). Cases of endometriosis lesions limited to the adipose layer had significant lower chance of using mesh ($p < 0.05$). However, adipose layer endometriosis lesions that had penetrated through the fascia layer and invaded into rectus abdominis muscle layer with/without peritoneum layer had significant higher chance of using mesh ($p < 0.05$). *Conclusions:* The more common position for scar endometriosis may be in the adipose layer at the corner of the surgical scar. Mesh therapy should be considered before surgery when the diameter of the abdominal wall mass detected by ultrasound is more than five cm and/or when the lesions invade into rectus abdominis muscle with/without peritoneum tissues from adipose and fascia layers.

Key words: Abdominal wall endometriosis; Surgical scar; Mesh; CA 125.

Introduction

Endometriosis is defined as functional endometrial glands and stroma occurring outside the uterus. The incidence of endometriosis in the abdominal wall is reported to be as high as 3.5% [1]. Many studies [2,3] have addressed the topic of abdominal wall endometriosis (AWE) since Nora *et al.* first reported 19 cases of AWE associated with cesarean section scars in 1956 [4]. In this retrospective study, the authors describe the exact anatomic locations of endometriosis in the abdominal wall, analyzing and discussing the difference between mesh and non-mesh groups, factors that may contribute to the AWE in the cesarean section surgical scars, and management of patients with AWE.

Materials and Methods

Following approval by the Anhui Medical University Review Board, a retrospective review was performed. Information of patients with AWE from January 2008 to December 2014 was searched from two large teaching hospitals (the first and second affiliated hospitals of Anhui Medical University) with a specific interest in gynecology. All patients had serum CA125 blood test and ultrasound with color Doppler imaging. The largest diameter of the lesion in the abdominal wall was determined by ultrasound examination. All masses proved to be scar endometriosis by

pathology after wide excision of the lesion under spinal anesthesia or general anesthesia with mask. The medical records of the patients including parity, age, time-gap between last surgery, onset of symptoms, previous surgeries, ultrasound examination, site of the AWE, initial diagnosis, cesarean section technique, definitive operation, complications, pathology reports, and recurrences were analyzed in detail.

Continuous variable are expressed as mean \pm standard deviation (SD). Chi-squared test was performed to compare incidences for each dichotomous variable. Statistical analysis was performed with SPSS 16.0. Difference was considered to be statistically significant when $p < 0.05$.

Results

A total of 95 patients with an age ranging from 26 to 48 years with a mean age of 33.5 ± 5.0 years at the time of excision were analyzed in this study (Table 1). During the same time from January 2008 to December 2014 in the two teaching hospitals, 16,972 cesarean sections had been performed. The occurrence rate of scar AWE in the study was 0.56%. Ninety patients (94.7%) had a history of one prior cesarean section and five patients (4.3%) had two prior cesarean sections. Seven patients also had a history of endometriosis ovary cystorectomy and another two patients had undergone a hysterectomy by laparotomy because of

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Table 1. — *Characteristic data of the patients.*

Characteristics	n=95 (%)
Maternity	
- Primipara	89 (93.4%)
- Multipara	6 (6.6%)
CA125 (U/ml)	
- Normal (<35 U/ml)	67 (70.5%)
- Elevated (ranged from 36.1~142.9 U/ml)	28 (29.5%)
Extent of lesions*, n=110 (%)	
- Invaded the adipose layer	102 (92.7%)
- Invaded the fascia layer	63 (57.3%)
- Invaded the rectus abdominis muscle layer	32 (29.1%)
- Invaded the peritoneum layer	13 (11.8%)
The time-gap, mean±SD (month)	
- Mean duration of symptoms, (ranged from 2~120 months)	28.01±25.21
- Cesarean section performed and the onset onset symptoms, (range 2~168 months)	32.76±29.18
- Previous cesarean section and the time of excision of scar endometriosis, (range 12~180 months)	60.99±36.10

* There are totally 110 abdominal endometriosis lesions among the 95 patients in this study.

uterine myomas. Eight-five patients (89.5%) had a gradually growing painful nodular abdominal mass in or adjacent to their cesarean incision scars, either non-cyclical ($n=18$, 21.2%) or cyclical ($n=67$, 78.8%) in nature. Six patients took analgesic drugs for the relief of severe pain. Only ten patients (10.5%) presented with a mass without pain.

Hard and fixed masses could be palpated near or in the scar position (Figure 1a). However the boundary of the mass was not clear in most of the cases, especially for the small nodules. All the patients underwent ultrasound with color Doppler imaging. The size of the abdominal wall masses varied from 0.5 to 7.0 cm, with an average of 3.25 cm confirmed by ultrasound. Most of the lesions near or in the scars were hypoechoic, vascular, and solid on ultrasound. Ten patients had undergone investigation by MRI due to atypical image in ultrasound. No additional diagnostic procedures such as fine needle aspiration cytology of the lesion under ultrasound guidance or core biopsy were performed in any case. Frozen section during operation was helpful in choosing the correct surgical procedure and no malignant tumor was proved by pathology during or after

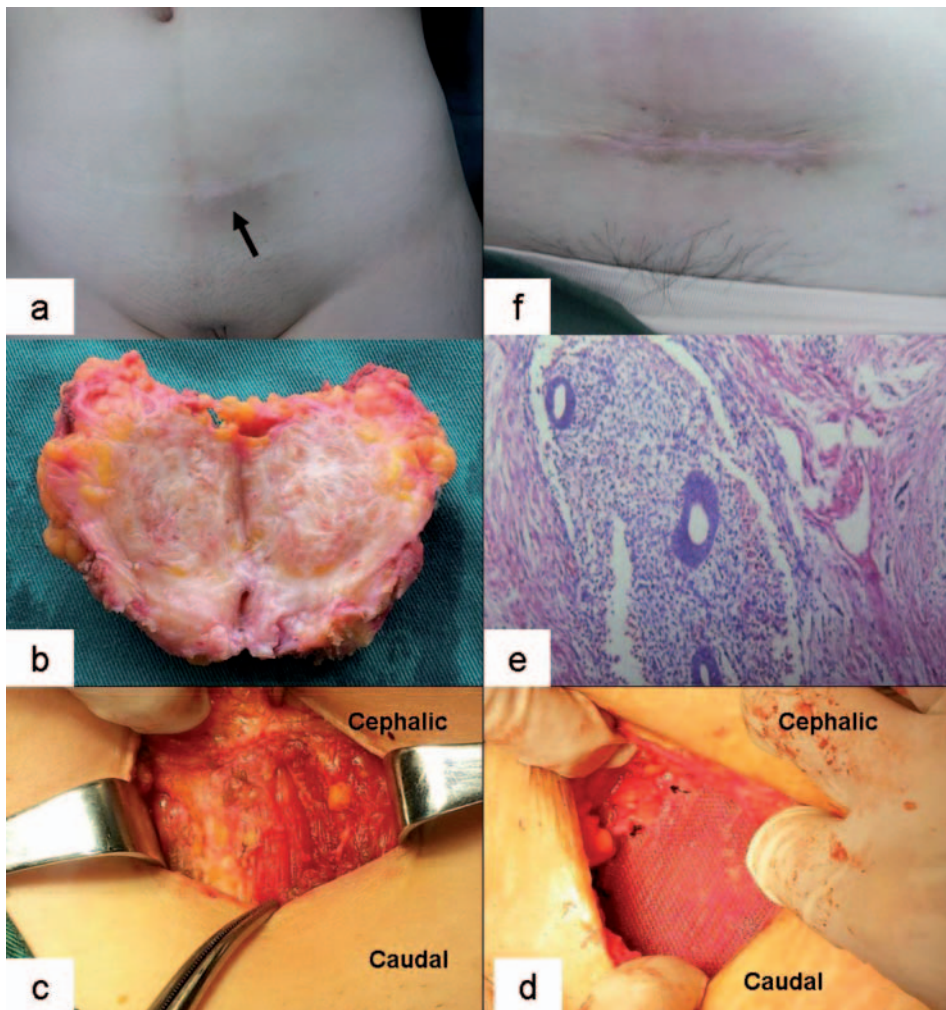


Figure 1. — A 36-year-old woman with an abdominal wall endometriosis mass after cesarean section.

- The skin with a transverse scar where endometriosis mass is located, indicated by a black arrow, is slightly elevated accompanied by pigmentation.
- Scar endometrioma invading into adipose, fascia, and part of rectus abdominis muscle which is excised with 1.0 cm of the normal tissue around the lesion.
- The incomplete fascia is difficult to close.
- Polypropylene mesh is fixed to the fascia by interrupted suture using non-absorbable thread.
- Haematoxylin and Eosin stained section of the abdominal wall endometrioma with endometrial glands.
- The incision recovered well at six-week follow-up.



Figure 2. — A 32-year-old woman presenting cyclical cyanotic changes in the subcutaneous tissue adjacent to the cesarean incision scars. An abdominal wall endometrioma is indicated by a white arrow and confined in the white circle could not be found without careful palpation.

the surgery even for one large mass reaching seven cm in diameter. One patient presented with cyclical cyanotic changes in the subcutaneous tissue adjacent to the cesarean incision scars (Figure 2). AWE lesion extending from the subcutaneous tissue to anterior wall of the uterus had been detected by ultrasound (Figure 3). The patient had both abdominal wall mass resection and uterus repairing procedure in one operation.

Following the imaging procedures, all patients underwent surgery. The surgery performed with/without mesh varied according to the site and size of the scar endometriosis (Table 2). Eighty-two patients complained of one mass (86.3%) in the abdominal wall, 11 patients (11.6%) each presented two lesions in the abdominal wall including ten patients with Pfannenstiel incisions, and one patient with midline incision, and two patients (2.1%) had three painful tubers in the abdominal wall, including one patient with Pfannenstiel incision and one patient with midline incision. Sixty-six patients (69.5%) with Pfannenstiel incisions had 78 lesions and 29 patients (30.5%) with midline incisions had 32 lesions after cesarean. For patients with Pfannenstiel incisions, the masses were found at the left, middle, and right of the cesarean incision scars were 31 (39.7%), 13 (16.7%), and 34 (43.6%), respectively, while in patients with midline incisions, the masses found at the upper, middle, and inferior portions of the cesarean incision scars were 12 (37.5%), seven (21.9%), and 13 (40.6%), respectively.

According to the position of the scar mass, about 1.0 cm of the normal tissue around the lesion was excised (Figure 1b). Polypropylene mesh was used if the incomplete fascia was difficult to close (Figures 1c, 1d). No special anti-adhesion mesh was required due to peritoneum defects in the study. A total of 18 patients had mesh therapy for fascia defect compared with 77 non-mesh therapy patients. The size of the lesions, mean duration of symptoms for painful mass, and level of the serum CA125 excluding seven patients with a history of endometriosis ovary cys-

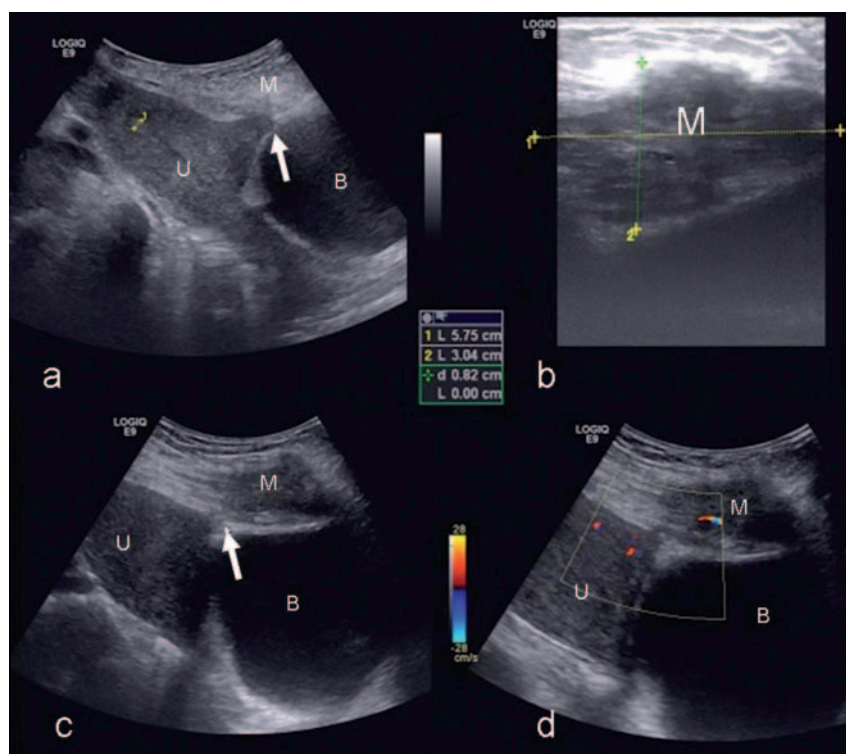


Figure 3. — The same patient mentioned in Figure 2. The abdominal wall endometriosis lesion extending from the subcutaneous tissue to anterior wall of the uterus is detected by ultrasound. Potential gap from the defect of the anterior uterus wall to the endometrioma in the abdominal wall is indicated by a white arrow. M: abdominal wall endometrioma; U: uterus; B: bladder.

Table 2. — *Clinical data between mesh and non-mesh use cases.*

	Non-mesh use case (n=77)	Mesh use case (n=18)	<i>p</i> value
Age, mean \pm SD (years)	32.93 \pm 5.03	34.88 \pm 4.55	0.09
Size of the lesion, mean \pm SD (cm)	2.60 \pm 1.10	4.97 \pm 1.09	0.00
Mean duration of symptoms of painful mass, mean \pm SD (months)	16.08 \pm 14.59 (n=70)	24.00 \pm 13.67 (n=15)	0.02
Cesarean section performed and the onset of symptom, mean \pm SD (months)	33.35 \pm 28.32	31.19 \pm 31.88	0.75
Previous cesarean section and the time of excision of scar endometriosis, mean \pm SD (months)	58.65 \pm 36.87	67.19 \pm 33.87	0.31
CA125 levels, mean \pm SD (U/ml)	26.46 \pm 19.41	55.69 \pm 36.78	0.00
Extent of lesions (n=110)*			
- Within the adipose layer with/without the skin layer, n (%)	46 (50.0%)	0	-
- Within both the adipose and fascia layers with/without the skin layer, n (%)	28 (30.4%)	4 (22.2%)	0.67
- Included the adipose, fascia and rectus abdominis muscle layers with/without the skin layer, n (%)	10 (10.9%)	6 (33.3%)	0.01
- Included the adipose, fascia, rectus abdominis muscle and peritoneum layers with/without the skin layer, n (%)	2 (2.2 %)	6 (33.3%)	0.00
- Invaded both the fascia and rectus abdominis muscle layers, n (%)	2 (2.2%)	1 (5.6%)	0.65
- Invaded both the rectus abdominis muscle and peritoneum layers, n (%)	1 (1.1%)	0	-
- Invaded the fascia, rectus abdominis muscle and peritoneum layers, n (%)	3 (3.3%)	1 (5.6%)	0.42

* There were a total of 110 abdominal endometriosis lesions among the 95 patients in this study, 92 (83.6%) in the non-mesh group and 18 (16.4%) in the mesh group.

torectomy (five in non-mesh used group and two in mesh group) were statistically different between mesh group and non-mesh group (Table 2). Cases for endometriosis lesions limited to the adipose layer had significant lower chance to use mesh ($p < 0.001$). However, adipose layer endometriosis lesions penetrating through the fascia layer and invaded into rectus abdominis muscle layer with/without peritoneum layer had a significantly higher chance to use mesh ($p < 0.05$) (Table 2).

The final pathological diagnosis for each lesion was AWE (Figure 1e). Most patients recovered well and symptoms that patients once complained of disappeared after surgery (Figure 1f). Only one patient presented with subcutaneous abscess after surgery and recovered well with drainage and antibiotic therapy.

Among the 95 patients, four patients were treated with gestrinone for three month before surgery and all reported a relief of severe pain and reduction of the mass; however, they all returned to the hospitals in six months due to the recurrence of pain and increasing mass after medication therapies. Two patients complained of one failed AWE surgery in other hospitals within two years and did not experience recurrence again after mass resection in the present hospital. Follow-up information covered three months to 5.5 years by telephone or clinic examination. Seventeen patients lost to follow up. Unfortunately, one patient with an AWE mass of four cm in diameter located in the adipose layer reported a recurrence mass for 1.0 cm in diameter by ultrasound with cyclical pain 18 months after the first surgery among the other 93 patients treated initially in the present affiliated hospitals. The patient received abdominal wall mass resection again and the lesion confirmed to be endometriosis mass by pathology. Three years later there was no other incidence

since the second surgery performed in the present hospital without medication therapy.

Discussion

AWE may be a more common extrapelvic endometriosis than reflected in the literature in women of reproductive age [5]. In the present study, the average age of the patients with AWE was 33 years (range 26-48), in agreement with other studies [6, 7]. The incidence of AWE after cesarean section in the present study parallels the results in the published literature and was between 0.03% and 1% [8-10]. All patients in this study had a history of at least one prior cesarean section and presented one to three abdominal wall masses that developed in a cesarean section scar. One patient even presented an AWE lesion extending from the subcutaneous tissue to anterior wall of the uterus. The opinion that cesarean section is a leading risk factor for the developing of AWE is well supported and a surgically-induced ectopic endometrial implantation theory is proposed [11, 12].

It is well-known that gradually growing nodular abdominal mass in or adjacent to their cesarean incision scars is an important symptom in the diagnosis of AWE. Ozel *et al.* [11] reported that 72.2% masses were found at the left margin of the Pfannenstiel incision scars, while Teng *et al.* [13] described 13 out of the 19 foci (68.4%) that were located in right corner of the Pfannenstiel incision wounds. The present study parallels the findings that endometrial inoculation after cesarean section easily present at both corner sites [14], but no predominant corner can be identified given that many doctors were involved in the study. Though there is no logical explanation why abdominal scar endometriosis

is more commonly present at the corner of the incision wound, some reports assume that they may due to the following: the endometrial cavity is cleaned with sponge after placental removal, uterus incision is closed and the abdominal wound includes the same suture material, abdominal wound is not thoroughly irrigated with saline solution before closing the abdominal wall, and the choice of surgeon's position may all contribute to the inoculum of endometriosis mass [13,15].

Ding *et al.* [16] described that the fascia was frequently affected whereas the peritoneum was least commonly involved with regards to invasion of the abdominal layers by AWE (36.1% VS 15.0%). Liu *et al.* [17] demonstrated the incidence of AWE invading the peritoneum reached 66.7% when resecting and repairing large abdominal wall incisions. In contrast to the aforementioned literature, the most common site to find endometriosis was located in the adipose layer of the abdominal wall as in the present study. This may be due to the fact that each study has its own objects which resulted in bias options. These studies also applied different statistical methods during their research. In the present study, the occurrence one adipose layer endometriosis lesion penetrated through the fascia layer and invaded into rectus abdominis muscle layer with/without peritoneum layer, which had an increased possibility for mesh grafts to be used in the surgery.

Although exact reason remains unknown, there is usually a significant time delay between the onset of symptoms and abdominal wall mass resection surgery [18]. Similar to others studies [13, 16, 18], the time between symptom onset and mass surgery was 28 months in the present study. The typical symptoms of AWE are the periodic painful mass or swelling associated with menses. It is difficult to diagnose AWE before surgery as patients complained of non-cyclic painful abdominal wall without a painful palpable or hard mass, which may explain the time delay between the onset of symptoms and the abdominal wall mass resection surgery in a part of the cases. All patients had abdominal wall masses in the present study, including 89.5% of gradually growing painful masses (78.8% cyclical vs. 21.2% non-cyclical) which in agreement with others studies [19]. Patients with large abdominal wall masses may endure long term painful symptoms than with small ones as in the present study. Ozel L *et al.* [11] assumed that as the mass grows and reaches a larger size, the pain becomes more cyclical in nature, but it was not confirmed by the present group.

The pathogenesis of endometriosis related pain is complex and manifold. Barcena *et al.* [20] assumed that the actual cause of the endometriosis related pain may be due to a result of a neurogenic inflammatory reaction besides the peritoneal lesions and adhesions. Research was intensified on the occurrence of endometriosis-related nerve fibres when large quantities of nerve fibres were detected in endometriosis lesions and infiltrated by stroma cells [21]. Vercellini *et al.* [22] mentioned that there exists a neuropathic

pain character as well as neurogenic inflammation processes besides the generally accepted inflammatory, nociceptive pain component, which was extremely particular when the initial cyclic pelvic pain becomes non-cyclic and/or chronic pain. The hypothesis merits further investigation for better understanding the pathogenesis of AWE related pain.

Owing to its practicality and low cost, ultrasonography is the most commonly used imaging examination performed to evaluate focal abdominal identified at physical examination. Images of AWE obtained by ultrasonography examination are non-specific in the literature. The typical ultrasound finding is a solid, non-homogeneous hypoechoic mass with spiculated margins infiltrating the surrounding tissue [10]. If ultrasound findings are inconclusive, MRI is suggested to determine the extent and nature of the focal lesions [23].

Fine-needle aspiration cytology is the initial approach for determining the nature of the mass and providing a diagnosis; however, its use remains challenging as it can cause abdominal cavity damage if an incision hernia had not been previously ruled out and has a potential diagnostic pitfall due to its rarity and occasional atypical cytological features [19, 24].

Although measurements of serum CA125 levels have been widely used in the gynecology field, the clinical significance of the serum CA125 elevation is still not fully understood in cases of endometriosis. The present study found that 7.4% of the patients with AWE had a history or subsequent diagnosis of pelvic endometriosis, which is within the range of the overall incidence of pelvic endometriosis in women of reproductive age [19]. This report also indicated that the serum CA125 level may increase in patients with large sizes of AWE. The present findings may offer some hints on the correlation between the size of the scar endometriosis and the value of the serum CA125 levels.

The malignant transformation of endometriosis in the abdominal wall is rare. No more than 30 cases have been reported in the literature [25]. The malignant type of endometriosis-associated neoplasm includes endometrioid carcinoma, sarcoma, and clear cell carcinoma. Malignancy transformation of endometriosis in the abdominal wall may occur in just a few months up to 18 years after surgery [10]. No malignant AWE were found among the present cases.

It is assumed in this report that medical therapy can temporary alleviation of symptoms, but cannot eliminate a mass, which is similar to that described by other authors [13, 14, 19]. A local wide excision of the scar endometriosis with at least one cm margin is recommended, although no studies have evaluated whether the surgical margin width affects the recurrence rate. The possible recurrence after surgery described in the literature reached 4.3% [19]. The present authors had less higher recurrence (1.1 %) due to adequate surgical excision, even resection of the muscle and fascia, and peritoneal elements of the

abdominal wall in order to obtain a good outcome. The mean diameter of the mass was 3.25 cm in the present series with an average of 4.97 cm in the mesh group by ultrasound. Based on the largest diameter of the lesions, Rodrich *et al.* [26] have classified tissue loss into small size (< five cm), intermediate size (five to 15 cm) and large size (> 15 cm) defects because the size of the defect has a major impact on the surgical therapy. Eltayeb *et al.* [27] mentioned in their study that cases with large ventral hernias (> four cm) could not obtain primary closure. The present authors' experience suggests that when the diameter of the abdominal wall mass determined by ultrasound examination is more than five cm larger, surgeons should be prepared for the possibility of abdominal wall defects and patients should be counseled that mesh repair may be necessary before surgical treatment.

Some suggestions for preventing AWE based on the implantation theory had been given in the literatures, such as using a wound edge protector to separate the edges of the incision, lifting the uterus outside of the pelvis before making the uterine incision, not using a sponge to clean the endometrial cavity, not using the same instruments and/or gloves which have touched endometrium, avoiding penetrating through endometrium layers when suture the uterine muscle, using separate needles for the abdominal and uterine closure, careful flushing and irrigating abdominal incision before closure, and extending the breastfeeding time to delay menstruation [12, 19, 20]. Above all, avoiding unnecessary cesarean section is recommended for preventing AWE.

Weakness of the present study included not using dimension as parameter to compare the difference between mesh and non-mesh use groups for short of information to calculate the exact size of the fascia defects during surgery. Other limitations included not comparing the relationships between the size and depth of the lesions and transverse and longitudinal incisions separately for having little mesh use cases in the present study. However, the present study provides some promising information for AWE diagnosis and therapy, but is limited by the fact that it is a single consultant retrospective non-randomised study. A case-control study should be used, the number of AWE cases needs to be increased, and more information should be collected in pre-, peri-, and postoperation in the future to authenticate the relationships between the size and depth of the AWE and the moment for mesh use.

Conclusion

AWE is a relatively common disease among women of reproductive age with a history of cesarean section and usually is underestimated by doctors and patients. The more common position for scar endometriosis may be in the adipose layer at the corner of the surgical scar. Periodic painful palpable scar mass with a cesarean section history strongly

suggests the diagnosis of AWE. Large size of scar endometriosis may accompany with higher serum CA125 level at the same time. Wide local excision with adequate margins is recommended for the treatment of AWE regardless if they are the primary or recurrent lesions. Mesh therapy should be considered before surgery when the diameter of the abdominal wall mass detected by ultrasound is more than five cm larger and/or the lesions invades into rectus abdominis muscle with/without peritoneum tissues from both adipose and fascia layers.

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