



Abdominal wall endometriosis: Case-series study and a systematic review

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Abstract

Abdominal wall endometriosis (AWE) is the presence of endometrial gland and stroma in the abdominal wall that should be kept in mind in differential diagnosis of pelvic pain due to especially increased C-section rates. Between January 2000 and July 2018, MEDLINE and EMBASE databases were systematically reviewed using the search criteria "abdominal wall endometriosis," "abdominal wall endometriomas,". Only the studies having over 20 patients were included. Case-series, case-control studies, and articles in languages other than English were excluded. Number of the patients, patients' age, study design, previous surgical history, most common symptom, time interval to symptoms, treatment, recurrence rate, and tumour size were investigated. In Total, 18 studies and 994 women were included in the study. Case studies, studies with less than 20 cases, non-English articles were excluded from the study. In the included studies, the numbers of minimum and maximum woman were 20 and 227, respectively. AWE significantly impairs the quality of life in reproductive age patients and is commonly seen in women with previous history of laparotomy, especially those who underwent cesarean section. Therefore, it must be kept in mind in the differential diagnosis of women who have a history of pain and a history of previous surgery.

Keywords: endometriosis, abdominal endometriosis, tumor, gynecology

1. Introduction

Endometriosis is a pathology defined as the presence of functional endometrial glands and stroma anywhere other than the uterine cavity (1). Although it is seen most commonly in pelvis, extrapelvic endometriosis is also observed with a frequency of 9-15% (2). Abdominal Wall endometriosis (AWE) is the most commonly seen extrapelvic endometriosis (3).

Abdominal wall endometriosis is defined as the presence of endometrial gland and stroma in the abdominal wall (4). In other words, it is the presence of endometrial tissue anywhere on the peritoneum. It is seen in 0.03% to 1.08% of patients undergoing obstetric or gynecological procedures especially hysterotomy (5). Three different theories have been proposed in the development of abdominal wall endometriosis. According to the direct implantation theory, endometrial cells scattered around abdominal wall during surgery, under the effect of hormones proliferate or cause metaplasia of the surrounding fascia. The second theory is the dissemination of endometrial cells by lymphatic or hematogenous routes (6). The third theory is that the pluripotent mesenchymal cells differentiate and form abdominal wall endometriosis (7). In addition, the anti-apoptotic surviving gene is thought to be effective in the formation and invasion of endometrial implants (8). Although AWE is a rare condition, it

significantly impairs the quality of life in women of reproductive age. In addition, AWE is now commonly seen due to the increased rates of cesarean section. Therefore, gynecologists, surgeons and radiologists should keep in mind this differential diagnosis in their daily practice.

In our study we aimed to present three cases of AWE who presented to our clinic. Most of previous studies on AWE were retrospective studies. Prospective studies are needed especially for diagnosis, treatment and follow-up. We have found that our results in this preliminary study are consistent with literature. We planned to do further prospective study regarding this topic.

2. Case Reports

2.1. Case 1

The patient was 33-year-old, G7P5L5 (gravida 7, parity 5, live birth 5). She had a history of four cesarean sections. The last cesarean section was five years ago. On superficial ultrasonography of the pelvic region at the right lower quadrant, lateral and to the right of the incision scar level, a poorly bounded heterogeneous hypoechoic, solid vascularized mass lesion seen with doppler, with a diameter of 24x11x15mm was observed on the posterior surface of the subcutaneous fat tissue. Surgical excision was performed. Histopathology was consistent with 4x2x2cm subcutaneous

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endometriosis. Within the first year no recurrence was observed in the patient.

2.2. Case 2

A 36 year-old patient G3P3L3 (gravida 3 parity 3 live birth 3) presented with pain and swelling in the left lower quadrant for the past one year. The patient had no history of operation other than bilateral tubal ligation with laparotomy 3 years ago. Abdominal ultrasonography revealed a mass with a size of 52x35x49mm, 5-6cm inferior to the umbilicus, in the left, antero-medial to the rectus with an oval, well-circumscribed hypoechoic homogenous, smooth echogenic septum. The mass was non-vascularized on doppler and was growing towards the subcutaneous fat tissue. Surgical excision was performed. Pathology results were consistent with endometriosis. No recurrence was observed in one year follow-up.

2.3. Case 3

A 40-year-old patient G2P2L2 (gravida 2, parity 2, live birth 2) presented with swelling in pelvic region for three months. The patient had two cesarean sections. She had her last operation four years ago. Abdominal ultrasonography revealed an irregular lesion with heterogeneous margins with a size of 22x8x19mm in the rectus abdominis muscle. Endometrioma was excised from the rectus muscle with laparotomy. Pathology results were consistent with endometriosis. The surgical margins were negative. There was no complication. No recurrence was observed within six-months of follow-up.

2.4. Systematic review methodology and results

MEDLINE and EMBASE databases were systematically reviewed. Between January 2000 and July 2018, search criteria were used as follows; "abdominal wall endometriosis," "abdominal wall endometriomas". The studies in which patients' numbers lower than 20 were excluded. Case-series, case-control studies, and articles in not English were also excluded. Therefore, in total, 18 studies were included (Fig. 1). Number of the patients, patients' age, study design, previous surgical history, most common symptom, time interval to symptoms, treatment, recurrence rate, and tumour size were investigated (Table 1).

3. Discussion

AWE is usually a pathology that may develop after surgical procedures. It is most seen after cesarean and tubal surgery. In the literature it has also been reported in patients without previous surgical history (9-11). Furthermore, it may also occur after laparoscopy, amniocentesis, episiotomy, and appendectomy (12-14). AWE is usually seen in women of reproductive age most commonly between 25-35 years (15-19). In 5.3% of the patients, it may be associated with pelvic endometriosis (20). Some risk factors for AWE were presented in the literature. AWE is more frequent in obese patients. The reason for this was thought to be the technical difficulty during the closure of the uterus (21).

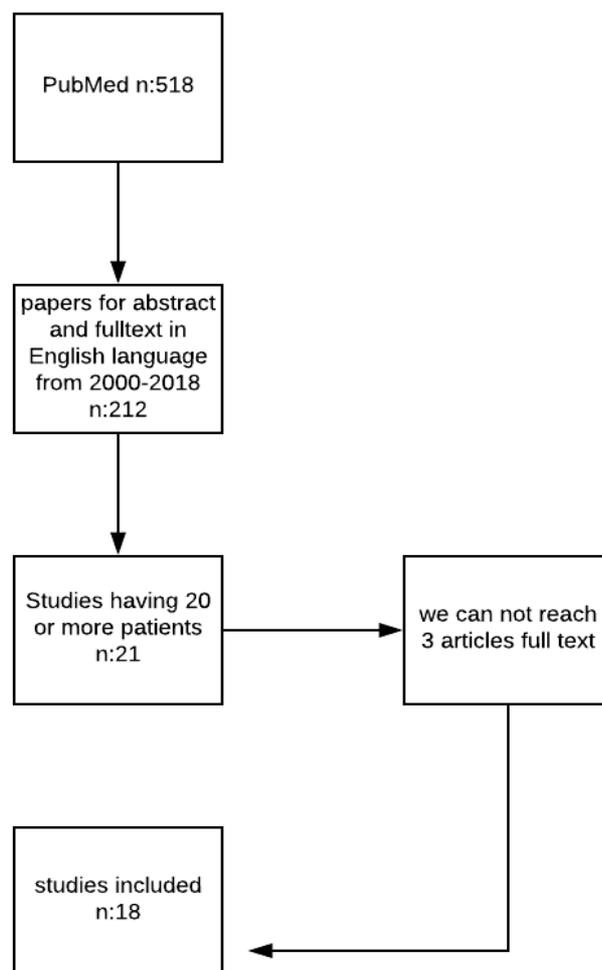


Fig. 1. Study flowchart

In addition, single part suturing of the uterus, not closing the parietal and visceral peritoneum during cesarean section are considered risk factors (22). The risk of AWE was found to be higher in patients with elective cesarean section than in patients who had undergone cesarean section following labor (23). High parity is thought to have a protective effect in AWE (24). Endometrioma is more common on the left side of cesarean section scar on the lower side of the midline incisions (20). The diagnosis of AWE can be easily made in the presence of the classic triad. History of cesarean section, increase in the intensity of pain during menstrual cycle, swelling around surgical scar (25). Although cyclic pain is characteristic for diagnose, noncyclic pain may be the presenting symptom and can cause misdiagnosis (26-30). Sometimes the presenting symptom may be only a palpable mass, dyspareunia, dysmenorrhea or cyclic hemorrhage from a superficial lesion (26). In order for the symptoms to occur AWE should reach a certain size. Onset of symptoms after first operation varies between 6 months to 20 years (14, 31-34).

Table 1. Results of studies

| | Year | Patient number | Age | Study Design | Most Common Prior Surgical History N (%) | Most Common Symptoms N (%) | Time Interval to Symptoms (months) | Treatment | Recurrence N (%) | Tumor Size cm (range) |
|------------|------|----------------|-------------------------------------|---------------|---|---|--|---|------------------|---|
| Kang | 2013 | 37 | 34 | Retrospective | Cesarean section 37 (100) | Palpable mass 36 (97.2) Pain 21(56.8) | 30 (Median) (6-96) range | WE 37 | 1 (2.7) | 3.5 (median) (1.0-10.0) |
| Bektas | 2010 | 40 | 32.3 | Retrospective | Cesarean section 36 (90) | Palpable mass 40(100) Noncyclic pain 18(45) cyclic pain 16(40) | 29.6 (Mean) (8-53) range | WE 40 | 3 (9.1) | 4.6 (mean) Range (3-6) |
| Teng | 2008 | 22 | 32.6 | Retrospective | Cesarean section 22 (100) | Palpable mass and cyclic pain 20 (90) Noncyclic pain with mass 2(10) | N/A | WE 22 | N/A | 4.4 mean (3-7) |
| Francica | 2009 | 28 | 31 | Retrospective | Cesarean section 28 (100) | Mass 27(96) Cyclic pain 22(78) Continuous pain 6(21) | 60 LSE 40 LSE | We28 | 0 (0%) | LSE 4.1 (3-6 cm) SSE 1.8 (0.7-2.6 cm) |
| Savelli | 2011 | 21 | 36 | Retrospective | Previous surgery for endometriosis 9 (25) Previous Cesarean section 8 (22) | Mass 18(86) Pain 19(91) | N/A | N/A | N/A | 2 (0,5-5) |
| Zhao | 2005 | 62 | 29,8 | Retrospective | Previous Cesarean section 61 (98) | Cyclic pain 56(90) Constant pain 5(8) Asymptomatic mass 3(5) | 28,6 (1-133) | WE 62 | 5 (8%) | 3,1(1-15) |
| Ecker | 2014 | 65 | 35 | Retrospective | Cesarean section 53 (81.5) | Abdominal pain 48(74) Mass/lump 41(63) | 48 (12-384) | WE 65 Laparoscopic 5 (7.7) Open 49 (75.4) Combined 11 (16.9) | N/A | N/A |
| Zhu | 2017 | 51 | 30.1 HIFU 31.4 Surgical Excision | Retrospective | Cesarean section 51 (100) | N/A | 24 (1-126) HIFU 17 (2-96) Excision | 23 HIFU 28 Surgical | N/A | 2,7 (1,3-6,8) HIFU 2,6 (0,7-5) Excision |
| Yela | 2017 | 52 | 30.7 1 | Retrospective | Cesarean section 34 (65.4) | Nodule pain 51(98) Mass 19(36) | N/A | N/A | 14 (26,9) | 3.98 |
| Khamechian | 2014 | 30 | 32,5 | Retrospective | Cesarean section 30 (100) | Mass 30(100) Pain (noncyclic) 10(33) Pain (cyclic) 14 (47) | 30.5 (2-53) | WE 30 | 1 | 2.59 (1-6) |
| Pas | 2017 | 71 | 34 | Prospective | Ceseraen section 71 (100) | N/A | 12 (1-168) | WE 71 | 1 | N/A |
| Maillot | 2017 | 20 | 30,1 | Retrospective | Cesarean 13 (65) | cyclic painful symptoms 19(95) Enlarged | N/A | We 13 Cryoablation 7 | N/A | 2.3 cm (0.5-7 cm) |

| | | | | | | nodules 6(30) | | | | |
|----------------|------|-----|-----------|---------------|---------------------------------|---|-----------------------|------------|--------------|-------------------------------|
| Ding | 2013 | 227 | 31,7 | Retrospective | Cesarean delivery 226 (99.6) | Abdominal mass 191(84) Cyclic pain 148 (65) Noncyclic pain 62 (27) | N/A | WE 227 | 3 (1.5%) | 2,9 (1–9 cm) |
| Francica | 2012 | 30 | 30.6 | Retrospective | Cesarean 30 (100) | Cyclic pain with mass 24(80) Contnuous pain 6(20) | 36 Median (12-120) | N/A | N/A | 2,7 (0,7-6) |
| Luo | 2017 | 32 | 39,4 | Retrospective | Cesarean 31 (96) | N/A | 11median (6-36) | 32 HIFU | N/A | 2,4 (1-5) |
| Khan | 2017 | 34 | 35.2 | Retrospective | Cesarean 30 (88.2%) | Abdominal pain 34(100). | N/A | WE 34 | 2 (5.9%) | 3,3 cm |
| zhang | 2016 | 151 | 31.2 7 | Retrospective | Cesarean 151 (100) | cyclic abdominal pain 121(80) irregular abdominal pain 17(11) | 31,4 (3-192) | 151 WE | 11 (7.3%) | 2,1 (1-6) |
| Wang Y | 2011 | 21 | 33.5 | Retrospective | Cesarean 20 (95) | cyclic abdominal pain 21(100) palpabl mass 21(100) | 10 (5-36) | 21 HIFU | N/A | 2,4 (range 1.0– 5.3 cm) |
| Ayşe at al. | 2018 | 3 | 36,3 | Retrospective | Cesarean 2 (67) | Cyclic pain 2(67) Palpabl mass 3(100) | 48 36-60 | WE 3 | 0 | 3,8 (2,2-5,2) |

The mean size of abdominal wall endometriosis varies between 2.3 and 3.2cm (35). Differential diagnosis includes tumor, hernia, lipoma, and hematoma. Ultrasonography is the first diagnostic tool. On abdominal ultrasonography, AWE shows deep pelvic endometriosis findings more than ovarian endometriosis findings (11). The ultrasound image is generally composed of a hyperechogenic ring showing edematous and inflammatory adipose tissue around a solid area (11).

CT and MR can be used in patients in whom a clear decision by ultrasonography cannot be made. Fine needle aspiration can sometimes be used for diagnosis. It is considered as a fast, reliable, and inexpensive diagnostic method (36). It can help in the diagnosis of malignant lesions. To make a diagnosis, at least two of the three criteria should be seen endometrial glandular cells, surrounding stromal cells and hemosiderin-laden macrophages (37). However, in this procedure there is a risk of spread of endometrial cells. For this reason, the needle tract should be removed during surgical excision. The histopathologic diagnosis of endometriosis is made in the presence of endometrial gland, stroma, and hemosiderin pigment (22).

Surgical excision used in the treatment is quite successful (16,20). During surgery, the nodule and the surrounding fascia should be removed so that there is a negative surgical

margin. Otherwise, there may be recurrence (10). The surgical margin should be 1cm (22). Therefore, sometimes broad facial defects may occur, and mesh may be required. Medical treatment is not preferred because of the recurrence of symptoms and low chance of success when the drug is stopped (10). Progesterone and GnRH analogues decrease the size of the lesions and decrease symptoms by suppressing menstruation (38). Ultrasonography-guided high-intensity focused ultrasound ablation (HIFU) is another method for treatment. Wang et al. described this method, but its long-term efficacy is not clear (17).

The risk of recurrence was reported to be 1.5 to 7.5% (20). The probability of progression of AWE to malignancy is reported as 1% (35). The most common malignancy is clear cell carcinoma. Besides, endometrioid, serous papillary, carcinosarcoma and mixed types are seen (38).

There are many recommendations for the prevention of AWE in the literature. Sumathy and colleagues suggested taking out the uterus while closing (22). Washing of the abdominopelvic space, bringing parietal and visceral peritoneum together have been suggested. The incidence of AWE increases when the same needle that is used to suture uterus is also used when closing abdominal wall (22).

Most of previous studies on AWE were retrospective studies. Prospective studies are needed especially for

diagnosis, treatment, and follow-up. We have found that our results in this preliminary study are consistent with the literature. We planned to do further prospective study on this subject.

To conclude, AWE, a pathology which frequency increases with time due to an increase in cesarean section rate, should be kept in mind in for differential diagnosis by general surgeons, gynecologists, and radiologists because they will often come across this disease. It is a problem that affects reproductive age patients, and it decreases comfort and impairs their quality of life

Conflict of interest

None to declare.

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None to declare.

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