



Abdominal wall mass suspected of endometriosis: clinical and pathologic features

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Objective

To evaluate the clinical and pathological characteristics of lower anterior abdominal wall masses suspicious for endometriosis.

Methods

A retrospective review of 38 patients who underwent surgery for a lower anterior abdominal wall mass suspicious for endometriosis was performed. Those with skin and intraperitoneal masses, lipomas, hernias, and metastatic malignant masses were excluded. Patient age, body mass index, delivery history, dysmenorrhea, and mass size and location were analyzed.

Results

Thirty-seven (97.3%) patients had a relevant surgical history, including 35 (92.1%) with a history of cesarean section (C/S). Among the three patients with no history of C/S, 1 underwent total abdominal and another total laparoscopic hysterectomy, and 1 had no previous surgical history. The mean (\pm standard deviation) size of the abdominal masses was 3.2 ± 1.2 cm. One patient developed a recurrent mass after excision of abdominal wall endometriosis. Trocar site endometrioma was found in one patient following total laparoscopic hysterectomy. According to the final pathology reports, endometriosis was found in 35 (92.1%) of patients. The remaining 3 patients (7.9%) had malignancy: adenocarcinoma, squamous cell carcinoma, and extra-gastrointestinal stromal tumor. Before surgery, only 3 patients (7.9%) underwent fine-needle aspiration biopsy of the masses, which were all postoperatively confirmed to be pathologically benign.

Conclusion

Although most abdominal wall masses in the present sample were endometriosis occurring at the scar site from a previous operation, 7.9% of patients ultimately exhibited malignancy. Therefore, all patients with suspected anterior wall endometriosis should undergo preoperative biopsy to identify the few that will have an alternative diagnosis.

Keywords: Endometriosis

Introduction

Endometriosis is characterized by the presence of stroma and endometrial glands outside the uterine cavity. This condition affects approximately 15% to 40% of women of childbearing age. The most common site of endometriosis is the abdominal cavity, specifically the pelvis, and occasionally, extra-pelvic sites [1]. Abdominal wall endometriosis (AWE) is a rare condition characterized by the presence of endometrial tissue in the subcutaneous fatty layer or muscles of the abdominal wall. AWE is usually caused by the dissemination

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of endometrial tissue at the incision site during obstetrical or gynecological surgery [2,3]. Malignant transformation of AWE is extremely rare. The most common malignant pathologies, reported in several studies, are clear cell or endometrioid carcinomas [1,4-6]. Endometriosis-associated abdominal wall cancers exhibit aggressive prognoses [4]. However, the estimated prevalence of AWE, especially malignant transformation of AWE, is no higher than 1%. Therefore, few investigations have examined AWE and malignant transformation, and most reports have been case studies or literature reviews [5,7,8].

In the present study, patients with clinically presumed AWE were evaluated. Pathological analyses of the abdominal wall masses made it possible to identify deficiencies in clinical procedures and make recommendations to improve management and prognosis.

Materials and methods

Eligible patients, seen in Bucheon, Seoul, Incheon, Uijeongbu at St. Mary's Hospital, St. Vincent Hospital, or the College of Medicine at The Catholic University of Korea (Seoul, Korea), were identified based on surgical history of suspected endometriosis associated with a lower anterior abdominal wall mass. Medical records between January 2010 and November 2017 were evaluated with regard to patient age, mass size and location, symptoms, and final permanent pathology following surgical resection. In all cases, the abdominal mass was situated between the umbilicus and symphysis pubis of the lower anterior abdominal wall. The inclusion criterion was a lower anterior abdominal wall mass suspicious for endometriosis before surgery. Patients with skin masses, intraperitoneal masses, lipomas, and hernias were excluded. Masses originating from a known malignancy (5 patients) were also excluded. Age, body mass index, delivery method and number, dysmenorrhea, menorrhagia, symptoms, mass size and location, and permanent pathology were analyzed. Cyclic pain of the abdominal mass, apart from previous dysmenorrhea, was analyzed separately. It was not possible to collect patient history of menorrhagia or dysmenorrhea among those who had previously undergone total hysterectomy. Abdominal wall mass size was reported according to long axis (cm).

Results

The mean (\pm standard deviation) patient age was 38.9 ± 6.7 years, and the mean abdominal mass size was 3.2 ± 1.2 cm (Table 1). Seventeen patients (44.7%) had dysmenorrhea, and 4 (10.5%) experienced pain from the abdominal mass during menstruation. Of 31 patients with available medical records, the interval from the most recent previous surgery to abdominal wall mass diagnosis was 5.93 ± 4.29 years. Although several patients complained of dysmenorrhea or menorrhagia before the latest surgery, endometriosis or endometrial cyst had not been confirmed at the previous surgeries. After discovery of the abdominal wall mass, 1 patient received gonadotrophin-releasing hormone agonist, and another took oral contraceptives to control pain and reduce the size of the mass. These treatments were ineffective; therefore, both patients ultimately underwent surgery. An ovarian endometrial cyst was found in 1 patient on magnetic resonance imaging (MRI) performed to evaluate the abdominal wall mass; this patient underwent laparoscopic ovarian cystectomy and abdominal wall mass removal.

Thirty-seven of the 38 patients (97.3%) had a history of surgery, including 35 (92.1%) who previously underwent cesarean section (C/S). Among the 3 patients with no history of C/S, 1 underwent total abdominal and another total laparo-

Table 1. Characteristics of patients with abdominal wall mass (n=38)

Characteristics	Values
Mean age (yr)	38.9 \pm 6.7 (36.6–41.1)
Mean BMI	22.9 \pm 4 (21.7–24.2)
Mean delivery number	1.6 \pm 0.7 (1.4–1.8)
Delivery mode	
Cesarean section delivery	35 (92.1)
Vaginal delivery	3 (7.9)
History of surgery	37 (97.3)
Cesarean section	35 (92.1)
Total hysterectomy	1 (2.6)
Total laparoscopic hysterectomy	1 (2.6)
Menorrhagia	10 (26.3)
Dysmenorrhea	17 (44.7)

Values are presented as mean \pm standard deviation (95% confidence interval) or number (%).

BMI, body mass index.

scopic hysterectomy, and 1 had no previous related/relevant surgical history. All 37 patients with a relevant surgical history exhibited a mass at the surgical scar.

The final pathology reports described endometriosis in 35 of the 38 patients (92.1%). Trocar site endometrioma was found in 1 patient with previous total laparoscopic hysterectomy. One (2.6%) patient experienced a recurrent mass 6 years after previous AWE excision (10 cm) that was larger than the initial mass (5 cm). A mesh was inserted during surgery for the recurrent mass. Due to incomplete medical records, it was not possible to collect information regarding resection margins from the pathology and surgical reports for all cases. Among the 35 patients, 25 (71.4%) did not attend outpatient clinic follow-up for >1 year.

Three of the 38 patients (7.9%) were diagnosed with malignancy: adenocarcinoma-originated endometrium or endocervix, squamous cell carcinoma, and extra-gastrointestinal stromal tumor (EGIST) (Table 2). The patient diagnosed with adenocarcinoma of the mass, who underwent a previous C/S,

was confirmed to have malignant transformation of the endometrium or endocervix. Her immunohistochemical results were ER(+), PAX8(+), GATA3(-), and WT-1(-). The patient diagnosed with squamous cell carcinoma had previously undergone total abdominal hysterectomy due to carcinoma *in situ* 20 years previously. The patient with no previous surgical history was pathologically confirmed with EGIST of the abdominal mass.

Thirty-seven patients underwent diagnostic imaging studies using computed tomography (CT) or MRI before abdominal surgery; however, no suspicious findings suggesting malignancy were observed on any scan. Before surgery, 3 patients (7.9%) underwent fine-needle aspiration biopsy (FNAB) of the abdominal wall mass, all of which were postoperatively confirmed to be pathologically benign. None of the patients with malignancy underwent presurgical biopsy.

Discussion

AWE accounts for 0.4% to 2.0% of patients diagnosed with endometriosis. It is found predominantly in the umbilical scar and scars from previous abdominal incisions, especially C/S, laparoscopy, or appendectomy [1]. In the present study, 34 of 35 patients with definitive AWE had a history of C/S; the other had undergone laparoscopic surgery. Their abdominal wall masses were situated at the scars associated with C/S or laparoscopic trocar sites. Several studies have reported port site endometriomas after laparoscopic surgery. Emre et al. [9] reported 15 cases and Siddiqui et al. [10] reported a case study of port site endometrioma. In the present study, the patient who had not undergone surgery had a final pathology report of EGIST. These data suggest that surgeons should consider the possibility of scar site endometrioma when operating. Ozel et al. [11] suggested that some C/S practices, such as certain bleeding control methods, abdominal cavity washing before closure, and minimizing subcutaneous dead space, may reduce the incidence of AWE. Surgical excision should include clear margins, at least 1 cm from solid tissue. In addition, medications such as gonadotrophin-releasing hormone agonist or danazol (a synthetic derivative of 17- α -ethinyl testosterone), both before and after surgical resection, may be effective in preventing AWE [12-14].

Malignant transformations of AWE are extremely rare, with clear cell carcinoma and adenocarcinomas reported in the

Table 2. Characteristics of abdominal wall mass (n=38)

Characteristics	Values
Presenting symptom	
Pain on abdominal mass not related to menstruation ^{a)}	1 (2.6)
Pain on abdominal mass related to menstruation ^{b)}	4 (10.5)
Palpable mass ^{c)}	25 (65.8)
a) + c)	1 (2.6)
b) + c)	7 (18.4)
Mass location	
Midline of abdomen	10 (26.3)
Right side	12 (31.6)
Left side	14 (36.8)
Bilateral	2 (5.3)
Subcutaneous	9 (23.7)
Subfascia	14 (36.8)
Muscle	15 (39.5)
Located in Cesarean section scar	35 (92.1)
Not located in Cesarean section scar	2 (5.3)
Pathology	
Endometriosis	35 (92.1)
Squamous cell carcinoma	1 (2.6)
Adenocarcinoma	1 (2.6)
Extragastrointestinal stromal tumor	1 (2.6)

Values are presented as number (%).

literature [4-7]. Taburiaux et al. [4] reported that 26 cases of malignant transformation of AWE were published between 1986 and 2014, among which the most common histological type was clear cell carcinoma, reported in 17 patients (65%). Other histological types in that report included endometrioid carcinoma in 6 patients (23%), serous papillary carcinoma in 2 (8%), and sarcoma in 1 (4%). Furthermore, de Almeida Gentile et al. [1] noted that malignant transformation involving the abdominal wall is extremely rare, with fewer than 30 cases of endometrioid carcinoma (70%), sarcoma (25%), and clear cell carcinoma (5%) reported worldwide. In the present study, 3 malignant abdominal masses were identified as adenocarcinoma of endometrial or endocervical origin, metastatic abdominal mass derived from cervical cancer, and EGIST, respectively. Most abdominal wall masses in females are benign; therefore, the malignancy rate reported in the present article is remarkable. Precancerous carcinoma *in situ* seldom recurs or metastasizes after total hysterectomy, especially as metastasis to the distant abdominal wall as cancer after a significant delay; therefore, we did not expect abdominal wall squamous cell carcinoma metastasis. Abdominal wall EGIST is also rare, with only a few cases reported [15,16]. After diagnosis of EGIST in the abdominal wall, we transferred this patient to the general surgery department.

In the present study, only 3 patients underwent FNAB before surgical treatment. In the study by Bektaş et al. [17], preoperative diagnosis of a lower anterior abdominal wall mass suspicious for endometriosis included 47.5% endometriosis, 20% abdominal wall tumor, 25% hernia, 5% desmoid tumor, and 2.5% suture granuloma. The abdominal wall mass was likely a tumor, although only 20% of patients underwent FNAB. Endometriosis was present in only 50% of those with a mass described in the final pathology report.

We analyzed the medical records of 28 of 35 patients who had not undergone FNAB, excluding 7 with incomplete medical records. The mean interval from original surgery to presentation of symptoms associated with the suspected AWE involving the abdominal wall mass was 14.1 months (range, 1–72 months). This delay in the treatment decision was due not only to delayed clinical visits by the patients, but also delayed decision-making by surgeons who overlooked possible malignancy. In the study by Horton et al. [18], this interval was 3.6 years.

According to several published studies, the incidence of AWE is low and mimics other clinical conditions, making pre-

operative diagnosis more difficult. Ultrasound, CT, and MRI are used to diagnose AWE [12,19]; however, these studies are not useful because they do not report significant findings. On ultrasound, AWE appears as a solid, heterogeneous hypoechoic mass with inner echogenic spots. The echogenic patterns depend on the hemorrhagic and fibrous components of the lesions. Although attenuation may vary, mild-to-moderate enhancement of the lesion has been observed on CT scans of the abdominal wall close to the C/S incision site [20]. In the present study, we did not detect any malignant masses on preoperative imaging. Therefore, we recommend FNAB for any abdominal mass suspicious for endometriosis before deciding on a treatment plan. According to Medeiros et al. [21], FNAB is a fast and accurate preoperative diagnostic method that prevents errors in approaching AWE. A history of confirmed pathological diagnoses may also improve patient prognosis. However, the use of FNAB is controversial due to the risk for new implants at puncture sites [12,22]. To avoid the spread of endometriosis after these procedures, it is advisable to include the aspiration site in the surgical resection field. Before performing FNAB, abdominal hernia should always be ruled out [23,24].

Several studies have reported the rare recurrence of AWE. Bektaş et al. [17] reported a higher rate of recurrence (9.1%) than in previous inadequate resection(s) of endometriotic lesions; another study reported a rate of 7.5%, and 1 review indicated a recurrence rate of 4.5% [24]. Many reports suggest that adequate surgical excision (1 cm) is essential for favorable outcomes [13]; however, to our knowledge, no studies have addressed the role of surgical margin size in recurrence. Although we have reported a patient with recurrent AWE, further studies are needed.

The present study was limited by the selection bias inherent to all retrospective designs, although this remains the best approach to investigating rare diseases such as AWE. Because we analyzed only 38 patients, statistical power may have been low. The major strength of the present study was that patients from 5 hospitals were included, increasing the generalizability of the results.

In conclusion, we found that most abdominal wall masses in patients whose histories we reviewed were benign endometrioses, and that all AWEs occurred at previous C/S sites. However, 7.9% of patients with a lower anterior abdominal wall mass suspicious for endometriosis turned out to have malignancy. Therefore, all patients with suspected AWE

should undergo preoperative biopsy to identify the few that will have an alternative diagnosis.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

Ethical approval

This study was approved by the Institutional Review Board (IRB No.: HC 19RESI0011) of St. Mary's Hospital.

Patient consent

This study approved the patient consent waiver due to the retrospective study design.

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