

Abdominal wall endometriosis: A monocentric continuous series and review of the literature

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Abstract

Introduction: Endometriosis is characterized by the presence of endometrial cells anywhere outside the uterine cavity. Endometriosis has been encountered on surgical scars especially resulting from a cesarean section recently. Our aim was to investigate the patients with abdominal wall endometriosis in our clinics where all the patients having this disease are managed via surgical resection. We also compared them with the patients presented in the literature.

Methods: A study was conducted on the patients with histopathological diagnosis of abdominal wall endometriosis as a case series. All the subjects were analyzed through a comprehensive medical assessment including documentation of the detailed history and physical and gynecological examination based on the patients' medical records.

Results: A total of 53 patients diagnosed with abdominal wall endometriosis were included in this study. The mean age and the mean body mass index of the patients were 32.6 ± 6.5 years and 25.2 ± 3.5 kg/m², respectively. A total of 49 patients had at least one operation history, mainly cesarean section. The most common sites for abdominal wall endometriosis were skin and the region containing the rectus abdominis muscle and rectus sheath, 56.6% and 18.9% respectively. The main symptom was the pain getting worse during menstruation and the presence of a mass in all patients. The main diagnostic tools in all patients were detailed history and physical examination, ultrasonography, and then histopathological confirmation after surgical excision.

Conclusion: Abdominal wall endometriosis is a common diagnosis mainly due to increased cesarean section rate. Clinical suspicion has the most important place in correct diagnosis and management of abdominal wall endometriosis.

Keywords

Extrapelvic endometriosis, scar endometriosis, abdominal wall endometriosis, vaginal endometriosis

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Introduction

Endometriosis is defined as the endometrial glands and stroma occurring outside the uterine cavity. Determination of the prevalence of endometriosis in the general population is difficult, because some women are asymptomatic and presentation of the women to the clinician shows great difference. However, this estrogen-dependent disorder occurs in approximately 10% of the reproductive-aged women and this rate increases up to 50% among infertile women.¹

Although the exact pathophysiological mechanism and the etiology of the endometriosis remain unclear, it is known that the pathogenesis of endometriosis appears to be multifactorial, including ectopic endometrial tissue, altered immunity, imbalanced cell proliferation and apoptosis, aberrant endocrine signaling, and genetic factors. The most widely accepted theory for the pathogenesis of endometriosis is the “ectopic endometrial implantation” theory named as Sampson’s theory of retrograde menstruation.²

Endometriosis can be classified regarding the localization as intrapelvic and extrapelvic endometriosis. Intrapelvic endometriosis defines the lesions in close proximity to the uterus. The most common sites are the ovaries, the fallopian tubes, the pelvic peritoneum, and the uterine ligaments, especially the uterosacral ligament and the broad ligament. In the extrapelvic endometriosis, the endometrial cells attach to any other location of the body including abdominal wall, thoracic cavity, brain, and musculature.³ The metastatic theory and the metaplastic theory are main theories explaining the development of extrapelvic endometriosis. The first one describes the implantation and the development of endometrial tissue at an abnormal location following surgical procedures such as cutaneous tissue. According to the metaplastic theory, endometrium directly arises from coelomic epithelium.

We aimed to present our experience about abdominal wall endometriosis, reported as the most common type of extrapelvic endometriosis in tertiary referral centers from Istanbul, including its diagnosis, association with the patients’ surgical history, management, and the clinical and demographic characteristics of the patients. We also compared our findings with those in the literature.

Materials and methods

This case series study was conducted on the patients with histopathological diagnosis of abdominal wall endometriosis, which is the most common type of extrapelvic endometriosis. We retrospectively analyzed 53 women at tertiary referral centers from Istanbul between January 2016 and June 2017. On histopathological examination, hematoxylin-eosin staining and Ki-67 staining had been used to confirm the diagnosis of abdominal wall endometriosis and any malignant transformation. We analyzed the patients’ medical records including all demographic and clinical

Table 1. Clinical and demographic characteristics of the patients.

Characteristics	Number (%) or mean \pm standard deviation
Age (years)	32.6 \pm 6.5
Body mass index (kg/m ²)	25.2 \pm 3.5
Parity	
0	3 (5.7)
≥ 1	50 (94.3)
Size (cm)	3.1 \pm 1.1
Comorbidity	
Absent	43 (81.1)
Present (hypertension, diabetes mellitus, hypothyroidism, and allergy)	10 (18.9)
Operation history	
Absent	4 (7.5)
Cesarean section	41 (77.4)
Cesarean section + laparotomy	8 (15.1)
Treatment for endometrioma	
Absent	42 (79.2)
Present	11 (20.8)
Location	
Incision	30 (56.6)
Incision + rectus abdominis muscle + rectus sheath	10 (18.9)
Rectus abdominis muscle	4 (7.5)
Umbilicus	4 (7.5)
Inguinal region	2 (3.8)
Episiotomy	2 (3.8)
Vaginal posterior wall	1 (1.9)
Mesh use	
Absent	47 (88.7)
Present	6 (11.3)

characteristics such as age, body mass index (BMI), number of gravida and parity, presence of comorbidity, and operation history. We also recorded the location, the size, the number, and the treatment of abdominal wall endometriosis. Our hospital’s Ethics Committee (Istanbul, Turkey) approved our study which was in accordance with the Declaration of Helsinki (diary number 2017-19/13). All procedures performed in the study were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all of the patients, allowing the use of their blinded clinical data for research purposes.

Statistical analysis

Statistical analyses were performed with the Statistical Package for the Social Sciences (SPSS Inc.; Chicago, IL, USA) Statistics 22.0 version for Windows. Means were presented with standard deviation (SD).



Figure 1. Macroscopic appearance of abdominal wall endometriosis, 305 mm × 348 mm (96 × 96 DPI).

Results

A total of 53 patients diagnosed with abdominal wall endometriosis were included in this study. All clinical and demographic characteristics of the patients are shown in Table 1. The mean age and the mean BMI of the patients were 32.6 ± 6.5 years and 25.2 ± 3.5 kg/m², respectively. In total, 94.3% of the patients were multiparous, with a range from 0 to 6 parity number. A total of 49 patients had at least one operation history, mainly cesarean section. There was no abdominal surgical history in only four women with extrapelvic endometriosis on episiotomy scars, on vaginal posterior wall, and on umbilical region. A total of 11 patients were receiving medical treatments including progestins, contraceptive pills, and progestin intrauterine device for endometriomas. Three patients had undergone laparoscopic endometrioma operation. The most common sites for abdominal wall endometriosis were the skin and the region containing the rectus abdominis muscle and rectus sheath, 56.6% and 18.9% respectively. The main symptom was the pain getting worse during menstruation and the presence of a mass in all patients. The mean diameter was 3.1 ± 1.1 cm with a range from 1.2 to 6.0 cm (Figure 1). There were a total of three recurrences. All the patients had been treated via surgical total excision and diagnosed with histopathological confirmation (Figures 2 and 3). The immunohistochemical staining with Ki-67 is shown in Figure 4. Only in one patient, the malignant transformation was observed as the

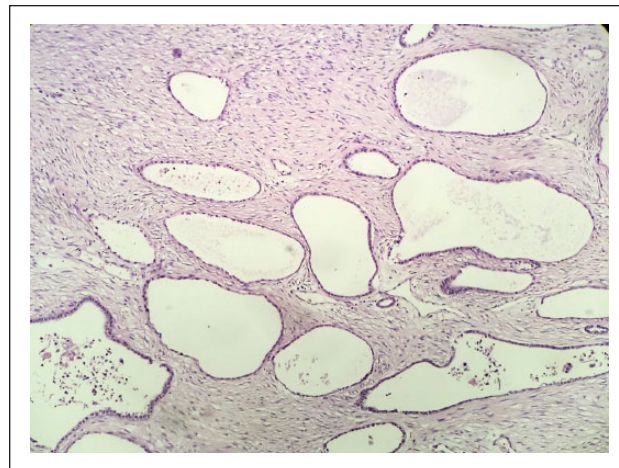


Figure 2. Histopathologic appearance of abdominal wall endometriosis with hematoxylin-eosin staining, 865 mm × 641 mm (72 × 72 DPI).

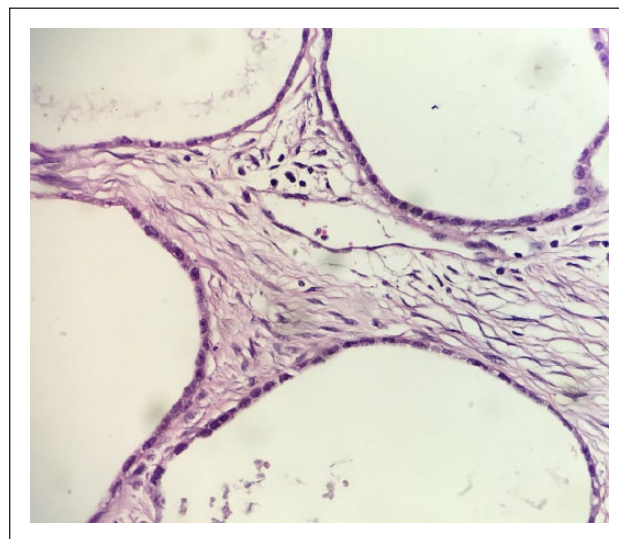


Figure 3. Histopathologic appearance of abdominal wall endometriosis with hematoxylin-eosin staining, 820 mm × 702 mm (72 × 72 DPI).

adenocarcinoma type (Figure 5). Endometriotic lesion located on abdominal wall had been excised during hysterectomy performed with the indication of myoma uteri in this 53-year-old woman. Immunohistochemical markers had been positively stained to confirm the malignant transformation. The lesions were single in 98.1% of cases, except only one case. She had two lesions with the mean diameter of 4.1 cm on the abdominal wall. The interval time between surgical intervention and the consultation of the patient with abdominal wall endometriosis was not recorded in all cases. The main diagnostic tools in all patients were detailed history and physical examination, ultrasonography, and then histopathological confirmation

with surgical excision. Due to the fascial defect, a prosthetic mesh had been applied to repair this defect in 11.3% of the patients.

Discussion

Endometriosis is described as the location of endometrial cells outside the uterine cavity in almost every area of the female body. Extrapelvic endometriosis is the occurrence of endometriosis outside the pelvic cavity, among them the most frequent location is the abdominal wall including

skin, rectus abdominis muscle, and rectus sheath.⁴ The prevalence of abdominal wall endometriosis is not exactly known, because of the irregular clinical picture, rarity of this entity, difficulty in differential diagnosis, and the consultation of the patients to the different specialists. Based on the small number of well-designed epidemiological studies and case series, the incidence varies between 0.03% and 0.4% in cesarean section scars.⁵ Abdominal wall endometriosis coexist in 12% of the patients with intrapelvic endometriosis.³ A total of 11 patients in our population were using medical treatment for associated endometriomas.

The pathophysiology of abdominal wall endometriosis could be explained with two theories mainly, the metastatic theory and the metaplastic theory. The first one, as the most widely accepted theory, describes the implantation and the development of endometrial tissue at an abnormal location following surgical procedures such as cutaneous tissue. According to the metaplastic theory, endometrium directly arises from coelomic epithelium. But there is no clear consensus on which theory best explains the different locations of abdominal wall endometriosis.⁶

Classifications have been made to facilitate the diagnosis and management of abdominal wall endometriosis based on the exact location and the size of the defect.⁷ But there is no uniformly accepted system for classifying abdominal wall endometriosis. It is known that abdominal wall endometriosis including skin, rectus abdominis muscle, and rectus sheath is the most common site for extrapelvic endometriosis. Cutaneous endometriosis is divided into two categories depending on its origin as primary and secondary. Primary cutaneous endometriosis is very rare and has no relationship

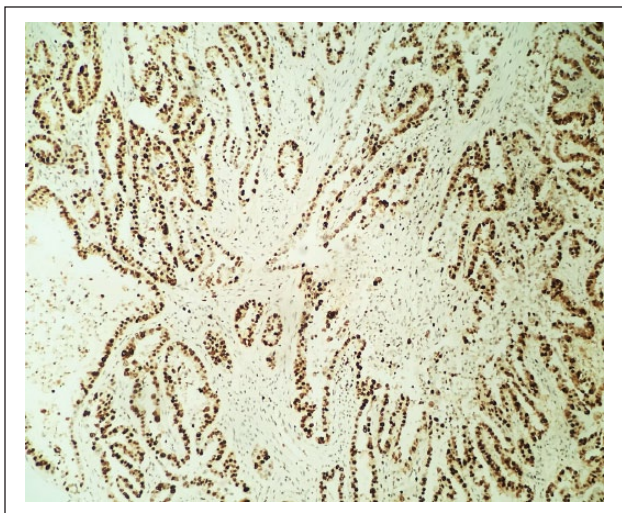


Figure 4. The immunohistochemical staining of abdominal wall endometriosis with Ki-67, 816 mm × 675 mm (72 × 72 DPI).

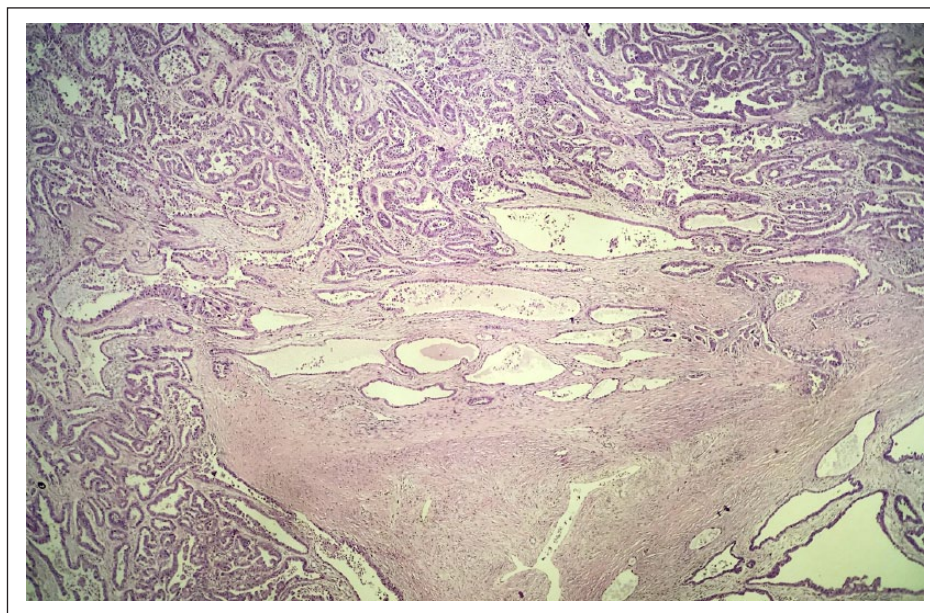


Figure 5. The malignant transformation in abdominal wall endometriosis as the adenocarcinoma type, 929 mm × 598 mm (72 × 72 DPI).

with prior abdominal or pelvic surgery. Because it is usually diagnosed in the umbilical region, it is also named as “umbilical endometriosis” or “Vilar’s nodule.”⁸ Secondary cutaneous endometriosis is associated with previous operations and accounts for more than 70% of the cases.⁹ Secondary cutaneous endometriosis, also named as scar or iatrogenic endometriosis, is observed most commonly on the scars of cesarean sections.¹⁰ The incidence of perineal endometriosis on the episiotomy has been reported as between 0.06% and 0.7% based on studies with a limited number of cases.³ The other locations in which extrapelvic endometriosis could be located are gastrointestinal tract, urinary tract, lungs, and muscular structures with neuronal involvement such as obturator internus muscle, trapezius muscle, deltoid muscle, and piriformis muscle.^{11,12} The most common location of extrapelvic intestinal endometriosis is the last part of the small intestine, the cecum and the appendix. However, there are cases reporting the diagnosis of extrapelvic endometriosis in the liver and gall bladder.¹³ Renal and urethral endometriosis are rarely encountered.¹⁴ Thoracic endometriosis is a rare entity including the presence of ectopic endometrial tissue in the pleura, pericardium, lung parenchyma, and rarely the diaphragm.³

In our centers, we experienced abdominal wall endometriosis on the cesarean scar site as the most common type of extrapelvic endometriosis, in accordance with the literature. A total of 49 patients had surgical history, mainly cesarean section among them. Only four patients had no prior operation, so these cases were primary abdominal wall endometriosis. The perineal endometriosis was encountered in two patients with previous vaginal birth history on the scar of episiotomy.

Depending on series, cutaneous endometriosis has been diagnosed in women with a mean age of 30–35 years. The mean age in our series was 32.6 ± 6.5 years ranging between 21 and 54 years, coinciding with the literature. The size of the lesion varies between 1 and 9 cm in previous publications. The mean size was 3.1 ± 1.1 cm ranging from 1.2 to 6 cm in our patients. It was reported that the interval time between the diagnosis of abdominal wall endometriosis and prior surgical intervention ranged from 1 to 20 years.⁹ We could not identify interval time in our series due to the lack of this information in our records.

The symptoms of abdominal wall endometriosis are often nonspecific and do not correlate with the severity or extent of the disease. Although it is not pathognomonic, symptoms are often cyclical in parallel with the menstruation period. The classic triad, including the pain associated with menstruation, presence of abdominal mass, and surgical history, is helpful in diagnosing the abdominal wall endometriosis.^{15,16} Our patients also consulted with the pain and a palpable mass swelling during menstruation period.

The differential diagnosis includes lipoma, hematoma, abscess, fasciitis, granuloma, and any malignant disease. Because of the difficulties in the diagnosis, the clinician

should consider the possibility of abdominal wall endometriosis when faced with any patient with the suspicious symptoms. A detailed history and physical examination are key for appropriate management. Imaging methods, especially ultrasonography and fine-needle aspiration cytology, are helpful in terms of diagnosis. Cystic, solid or mixed lesions, and hypoechoic nodules infiltrating the surrounding tissue are observed on ultrasonographic images. Ultrasound-guided biopsy and fine-needle aspiration cytology are fast and accurate methods for the diagnosis avoiding errors before management of abdominal wall endometriosis.¹⁰ Magnetic resonance imaging (MRI) distinguishes the different planes between subcutaneous tissue and the abdominal muscle. However, all these diagnostic methods are not practical. Biochemical markers such as CA-125 could be used in women with symptoms suggestive of abdominal wall endometriosis. However, it is not specific and well correlated with the lesion. The best method for both definitive diagnosis and treatment is wide surgical excision which also enables to rule out malignancy. Histopathological examination shows the characteristic endometrial glands and stroma, usually accompanied by a chronic inflammation and hemorrhage. Immunohistochemical markers such as CD10, Ki-67, cyclooxygenase-2, and vascular endothelial growth factor are increasingly being used to confirm the diagnosis.⁹ Mesh replacement may be required due to the fascial defect after wide excision of the lesion infiltrating the rectus sheath beneath the skin.

In our series, history, physical examination, and ultrasonography had essential roles in detecting and localizing abdominal wall endometriosis. Surgical removal of all visible endometriotic implants with safe margins had been applied in all patients. Immunohistochemical staining with Ki-67 was used for differential diagnosis. We placed prolen mesh in six patients providing the fascial integrity.

Drug therapy such as progestins, danazol, contraceptive pills, and gonadotropin-releasing hormone agonists provide temporary relief of pain and the other symptoms, but they usually recur in time.¹⁷ These hormonal treatments may be preferred for reducing the size of the lesions before the surgery. In total, 20.8% of our patients were using hormonal treatments for associated endometriomas diagnosed with ultrasonography. Sclerotherapy, chemical cauterization with silver nitrate, laser vaporization, and percutaneous cryocoagulation are the alternative treatment methods, all of which controversial options to abdominal wall endometriosis, because both medical treatment and alternative methods have no ability to diagnose abdominal wall endometriosis and to rule out malignant disease.¹³

Although a rare event, abdominal wall endometriosis may show malignant transformation. Endometrioid or clear-cell carcinoma, adenocarcinoma, and sarcoma have been described to develop in relation to abdominal

wall endometriosis based on case reports with the risk of 0.3%–1%.⁹ Because of this possibility, wide excision with at least 1-cm margin is proposed to prevent residual endometriotic tissue.¹⁴ The histopathological examination had confirmed the adenocarcinoma type of malignant transformation in one patient in our population. This woman had undergone hysterectomy due to the presence of multiple myomas. She had the pain and a palpable mass swelling during menstruation period. This endometriotic nodule had been excised following hysterectomy during the same operation. Malignant transformation was shown with immunohistochemical markers.

There is a little recurrence risk usually during the year after surgery in between 4% and 11%.⁹ In our study, three patients had been operated for the second time in a year after the first operation due to the recurrence of abdominal wall endometriosis on the same location.

Several preventive intraoperative measures have been proposed to minimize the iatrogenic implantation of endometrial tissue. Using different suture materials in the uterus and abdominal wall, minimizing the contact of the compresses used for swabbing the endometrial cavity, and cleaning the surgical area with physiological saline before closure can be helpful in preventing its development, although there are no well-designed studies evaluating the effectiveness of these measures.

To our knowledge, this study has the largest sample size performed in our population about the abdominal wall endometriosis. It is also a strength that our study emphasizes the importance of malignant transformation of abdominal wall endometriosis.

This study has several limitations. First, our study was performed with a retrospective design. Second, the incidence rates can be underestimated due to the presence of asymptomatic cases. Only women with surgically proven abdominal wall endometriosis were included in our study.

As a conclusion, clinical suspicion has the most important place in the correct diagnosis and management of abdominal wall endometriosis. All clinicians should have more awareness of this condition. It is important to be careful and take precautionary measures to prevent abdominal wall endometriosis, which may be more frequently encountered in parallel with the increasing cesarean rate recently.

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Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Our hospital's Ethics Committee (Istanbul, Turkey) approved our study, which was in accordance with the Declaration of Helsinki (diary number 2017-19/13).

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Informed consent

Written informed consent was obtained from all the subjects for participation before the study and for publishing their anonymized information in this article.

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