Clinical and Surgical Characteristics of Abdominal Wall Endometriosis: A Multicenter Case Series of 80 Women

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Abstract. *Background/Aim: Endometriosis of the abdominal wall (AWE) is poorly understood because of its rarity and heterogeneous nature. The aim of this study was to investigate and present the clinical and surgical characteristics of AWE and to propose its classification. Patients and Methods: This was a multicentric retrospective study. For this analysis, the data from three endometriosis centers were collected. In total 80 patients were included in this study. The Academic Hospital Cologne Weyertal is a certified, level III endometriosis center in Germany with 750-1,000 endometriosis surgeries being performed annually; Barzilai University Medical Center is a certified endometriosis center in Ashkelon, Israel; and Baku Health Center is an endometriosis Center in Baku, Azerbaijan. Results: The size of nodule (histological specimen) was significant larger in women with than those without adenomyosis (3.34±1.4 vs. 2.55±1.33 cm, p=0.016). The incidence of subfascial involvement was also found to be significantly higher in these women (42% vs. 19%, p=0.03). No significant difference was found in patients with and without obesity. In 78% of cases, the proliferation level (Ki67 marker) was less than 30%. Conclusion: AWE has a high prevalence of symptoms such as abdominal wall pain and swelling, as well as bleeding. The strengths of the current study are the investigation of the proliferation marker Ki67 in AWE, the impact of adenomyosis, as well as the suggested classification.*

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Endometriosis of the abdominal wall (AWE) is one of the rarest forms of the disease, with a reported incidence of 0.03-3.5% (1, 2). It is often either misdiagnosed as a hernia, hematoma, or lipoma, or neglected, since patients are usually referred to general surgeons, and is poorly understood by gynecologists because of its heterogeneous nature (2). AWE is most likely an aggregation of etiopathogenically different conditions unified by one common feature – the implantation and embedding of ectopic endometrial tissue within the same anatomical area (1). Frequently described localizations are cesarean section scars, followed by laparoscopic trocar tracts and the navel (2, 3). Moreover, rising occurrence in recent times his been linked to increased surgical activity in obstetrics and gynecology, and mainly to abdominal delivery, thereby strengthening the general opinion on the mechanical origin of AWE (1). However, if surgical violation of the integrity of biological layers (*i.e.*, incision) with mechanical spreading of endometrial cells (hysterotomy) is assumed to be a major pre-condition for its development, it is possible to divide AWE into rare spontaneous or scarless (umbilicus, inguinal canal) forms and frequent postsurgical (scars from cesarean section or trocar insertion) variations (3, 4). Patients with AWE can have a variety of symptoms including cyclical abdominal pain, or a palpable nodule, as well as swelling. In addition, typical complaints of endometriosis such as dysmenorrhea, dyschezia, dysuria or dyspareunia, can also occur (5).

Some authors described a clinical triad for suspicion of AWE: A history of open gynecological surgery, palpable abdominal mass and catamenial pain (6). Traditionally, clinical evaluation in such cases requires ultrasound, magnetic resonance imaging (MRI) or with both subsequent surgery and histopathological examination to make a final diagnosis (1, 7, 8). No existing classification systems mention AWE alongside other forms of endometrioid disease, except for the #ENZIAN, which simply classifies it as "F (localization)" (9).

For curative treatment, complete local surgical excision is required (3). As in case of intrapelvic or intrabdominal implantation, the depth of AWE might be used to distinguish the severity of the disease, which is of particular importance for proper management and better outcomes.

The goal of this study was to investigate and present the clinical and surgical characteristics of AWE. Furthermore, the authors of this paper suggest a classification of AWE.

Patients and Methods

This was a multicentric retrospective study; according to 15§ of the professional code of the North Rhine Medical Association, neither advice nor an ethics vote is necessary for a retrospective study.

For this analysis, the data from three endometriosis centers was collected. The Academic Hospital Cologne Weyertal is a certified level III endometriosis center in Germany, undertaking 750-1,000 endometriosis surgeries annually; Barzilai University Medical Center is a certified endometriosis center in Ashkelon, Israel; and Baku Health Center is an endometriosis Center in Baku, Azerbaijan. In total, 80 cases with AWE were included, with 51, 16 and 13 cases, respectively. Only patients with AWE symptoms and who underwent surgery were included in this analysis. Histological confirmation of the resected endometriosis nodules was mandatory. Preoperative assessment was performed by abdominal ultrasound and by MRI. In cases when the patients not only had symptoms related to AWE, but also those common to endometriosis, such as dysmenorrhea, dyschezia, dysuria or dyspareunia, a laparoscopy was also performed. Endometriosis was classified according to the #ENZIAN classification (9). In the majority of cases at the Academic Hospital Cologne Weyertal, the proliferation marker Ki67 was examined. Patients were divided into three groups based on the percentage of Ki67 expression: below 10% was considered low, 10-30% intermediate, and over 30% was considered high.

In this analysis, the ultrasound and MRI findings for the preoperative assessment were compared. Furthermore, the impact of adenomyosis and the number of cesarean sections in association with AWE were investigated. Adenomyosis was diagnosed using transvaginal ultrasound or during laparoscopy. For the ultrasound assessment, the Morphological Uterus Sonographic Assessment criteria were taken into account (10). The correlation between body mass index (BMI) and AWE was also examined. The involvement of the fascia was also evaluated in each case. Depending on the depth of endometriosis infiltration and involvement of the fascia, AWE was defined into three types: Epifascial (subcutaneous ± fascia), subfascial (rectus muscle ± fascia) and total infiltration (subcutaneous, fascia, as well as the rectus muscle). The localization of AWE in relation to the abdominal midline was defined as left, right or center.

Statistical analyses were carried out using Fisher's exact test, with descriptive statistics and confidence interval of the mean, as well as *t*-test to compare two means. The data are given as the mean and standard deviation

Results

We retrospectively evaluated women who underwent surgical excision of AWE at three tertiary care centers. In total, 80 women with histological confirmation of endometriosis were enrolled in this study. Figure 1 shows some examples of the AWE cases of the present study.

The patient demographic characteristics are shown in Table I. The mean age of the patients was 36±5.4 years. The mean BMI was 26.19±5.23 kg/m^2. Obesity (BMI ≥30) was observed in 17 (21.25%) women. Nulliparity was observed in 25% of the women, whereas 70% of women had a history of cesarean delivery.

Table II presents clinical features related to endometriosis. Abdominal wall pain was the leading symptom related to endometriotic nodules and was observed in 69 women (86.25%). Bleeding of endometriotic nodules during the menstrual cycle was observed in 14 women. In 13 cases with cyclic bleeding, the localization of the nodule was in the umbilicus.

Endometriotic nodules were commonly observed in patients with a cesarean scar (65%). The mean number of cesarean sections in women with scar endometriosis was 1.61±0.75. In two women with a previous cesarean section, the endometriotic nodule was observed on the abdominal wall far from cesarean scar and the umbilicus. The vertical distance between the nodule and the cesarean scar was 5 and 6 cm, respectively. As shown in Table II, these two cases were defined as "other localization". Trocar site endometriosis was observed in four women. All of them had undergone previous laparoscopic surgery due to endometriosis and all endometriotic nodules were observed on the left accessory trocar tract. Data about the location of the nodules in relation to the abdominal midline was available for 58 women (52: cesarean scar; 4: trocar site; 2: other localization). Left side dominancy was observed (Table II). In 22 cases, the nodule was in the umbilicus, so side differentiation was not possible.

Ultrasonography was the common diagnostic imaging modality and performed in 87.5% of cases. The mean nodule size that was measured histologically was larger than the preoperative size measured by both MRI and ultrasonography (2.79±1.37 *vs*. 2.22±1.21 cm).

Clinical features of umbilical endometriosis. In 22 women, endometriotic nodules were observed in the umbilicus. The incidence of cyclical bleeding from the nodule was 60% (n=13) and was observed more commonly than cyclical bleeding from nodules in other locations (17.5%; Table II). No significant relationship was observed between umbilical
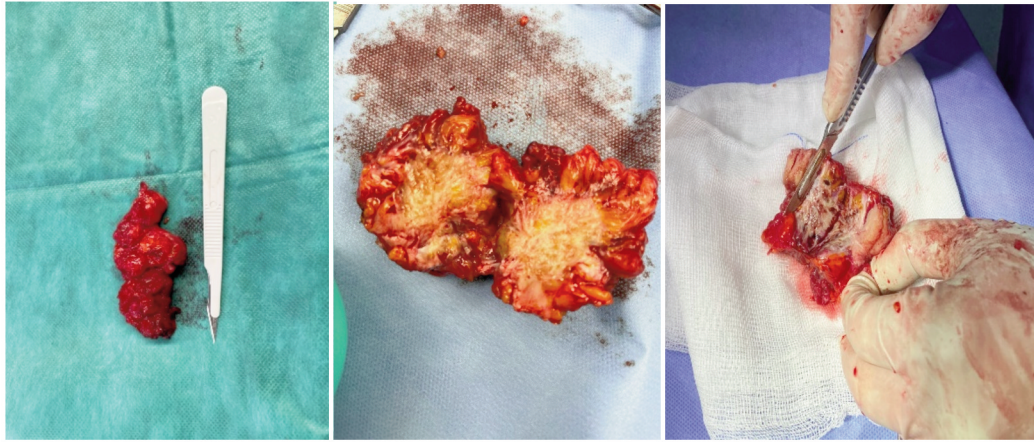
Figure 1. *Postoperative imaging of resected abdominal wall endometriosis with internal fibrotic changes.*

Table I. *Patient demographic characteristics (n=80).*

Characteristic	Value
Age, years	
Mean±SD	36±5.4
BMI, kg/m²	
Mean±SD	26.19±5.23
Obstetrical history, n (%)	
Nulliparity	16 (20%)
Vaginal deliveries	20 (25%)
Cesarean deliveries	56 (70%)
Number of cesarean deliveries, n (%)	
1	29 (36, 2%)
2	22 (27.5%)
≥3	5 (6.2%)
Previous gynecological surgeries, n (%)	
Myomectomy	3 (3.75%)
Hysterectomy	5 (6.25%)
Laparoscopy (adhesiolysis, adnexal)	22 (27.5%)
Preoperative hormonal treatment, n (%)	
Yes	17 (21.25%)

BMI: Body mass index.

endometriosis and a history of cesarean deliveries. Of the 22 women with umbilical endometriosis, only two (9%) had had a previous cesarean section; 15 women (68%) were nulliparous. In 18 women (82%), either pelvic endometriosis or adenomyosis was observed. However, only seven women (32%) had had previous laparoscopic surgery with excision of endometriosis. In all cases with umbilical endometriosis, the nodule was epifascial; however, considering the whole study population, the rate of epifascial lesions was 72.5% (Table II).

The relationship between adenomyosis and clinical features of AWE. Of the 80 women, adenomyosis was found in 26 (32.5%). Table III shows the comparison of the clinical characteristics between women with and without adenomyosis. The size of the nodule (histological specimen) was larger in women with adenomyosis than in those without, with the statistically significant difference (3.34±1.4 *vs*. 2.55±1.33 cm, *p*=0.016). The incidence of subfascial involvement was found to be significantly higher in women with adenomyosis than in those without (42% *vs*. 19%, *p*=0.03).

Table IV shows the relationships between the BMI and clinical features. No significant difference was found between patients with and without obesity.

Discussion

Our series of AWE included cesarean scar, umbilical and previous trocar locations. The analysis of its clinical and pathological characteristics allows us to draw some conclusions and make proposals concerning its diagnosis and treatment.

There is a significant number of publications devoted to abdominal endometriosis but most of them are case reports or case series with accompanying review. Partly because of this, most of them focused on the location and the possible route of origin of the implants (4, 11, 12). Previous reports define abdominal endometriosis in different localizations, including scar, umbilical, and inguinal (4, 11, 12). In the current series, we did not observe any cases of inguinal endometriosis. Due to the possible etiology of peritoneal fluid spread via the round ligaments and the assumable direct relationship with pelvic endometriosis (13, 14), inguinal endometriosis can be considered an extension of pelvic endometriosis into the inguinal canal and should not be included as a type of AWE. Despite many previous reports, in the literature there is lack of acceptable classification

Table II. *Clinicopathological characteristics related to endometriosis (n=80).*

Characteristic	Mean±SD; n (%)
Symptoms	
Local	
Abdominal wall pain	70 (87.5%)
Swelling of nodule	31 (38.7)
Nodule with cyclic bleeding	14 (17.5%)
General	
Dysmenorrhea	47 (58.7%)
Dyspareunia	13 (16.25%)
Dyschezia	3 (3.75%)
Localization	
Cesarean scar	52 (65%)
Umbilical	22 (27.5%)
Trocar site	4 (5%)
Other	2 (2.5%)
Side	
Left	38 (66%)
Right	18 (31%)
Middle	2 (3.4%)
Diagnostic method	
Sonography	70 (87.5%)
Magnetic resonance	10 (12.5%)
Size of nodule, mean±SD, cm	
Imaging	2.22±1.21
Histological	2.79±1.37
Size on imaging *vs.* histology, mean±SD, cm	
Sonography	2.24±1.25 *vs.* 2.77±1.4
MRI	1.94±0.77 *vs.* 2.87±1.02
Ki67 (N=33)	
<10%	19 (57%)
10-30%	7 (21%)
>30%	7 (21%)
Fascial involvement, n (%)	
Yes	42 (52.5%)
Extension of the disease	
Epifascial	58 (72.5%)
Subfascial/total*	22 (27.5%)
Other endometriotic lesions, #ENZIAN classification	
P	32 (40%)
O	10 (12.5%)
A	9 (11.2%)
B	18 (22.5%)
C	6 (7.5%)
FA	26 (32.5%)
FB	2 (2.5%)
FI	5 (6.2%)

MRI: Magnetic resonance imaging; SD: standard deviation. *Total infiltration of abdominal wall including rectus muscle, fascia, and subcutaneous tissue. #ENZIAN classification: P: peritoneal; O: ovarian; A: vagina; rectovaginal space; B: uterosacral ligaments/cardinal ligaments/pelvic sidewall; C: rectal; FA: adenomyosis; FB: bladder; FI: other intestinal locations (sigmoidal colon; small bowel).

regarding clinicopathological features of AWE. In the current study, we defined AWE into the following categories: cesarean scar, accessory trocar, umbilical, and other. We consider the depth of the lesion the same as for deeply infiltrating endometriosis and to be of greater clinical importance; we therefore propose the fascia as a threshold to distinguish the degree of invasion. Table V shows the new classification of AWE we suggest, with common clinical features that were observed in this analysis. This classification might be helpful regarding clinical evaluation, intraoperative expectations, and postsurgical outcomes.

Another important finding concerns dysmenorrhea as a presenting symptom of pelvic endometriosis, which was revealed in 60% of our patients. This may be attributed to coexistence of pelvic endometriosis observed in up to 40% of our series, which is higher than previously reported of 6-13% (4, 15). The reason for this difference can be explained by the additional laparoscopic evaluation, which revealed peritoneal endometriosis in 40% of the study population (Table II).

In the current series, umbilical endometriosis was found to be the second most common type of AWE (Table I). Twenty-two women presented with umbilical endometriosis (27.5%). Nine women (41%) had a history of previous abdominal surgery; there is a lack of data about the presence of umbilical endometriosis before surgery in these nine cases. However, in 13 women (59%), there was no history of abdominal surgery, making these cases of primary umbilical endometriosis. Primary umbilical endometriosis was described for the first time in 1886 by Villar (16, 17). The primary form of umbilical endometriosis is the most common, comprising 69% of umbilical endometriosis (18). In our series, 82% of women with umbilical endometriosis also presented documented pelvic endometriosis or adenomyosis. The co-existence of umbilical and pelvic endometriosis might be explained by several plausible theories, such as the metastatic hypothesis, genetic predisposition or via embryological remnants (19, 20).

Furthermore, in our series, 32.5% of women had also been diagnosed with adenomyosis. In those women, the size of AWE was found to be larger than in the women without adenomyosis. Our results revealed that the coexistence of adenomyosis may be a sign of more extensive AWE, with larger nodules and deep infiltration of the abdominal wall (Table I). This insight may be useful for surgeons in terms of appropriate presurgical evaluation. In contrast to adenomyosis, no significant difference was found related to BMI (Table IV).

The most common localization of AWE is a cesarean scar (2). However, the risk of AWE does not depend on the number of cesarean sections; 29 out of 52 patients with cesarean section scar endometriosis (55.7%) had only one cesarean section in their medical history (Table II).

The common acceptable treatment of choice for AWE is the total surgical excision of nodules. The pooled recurrence

Table III. *The relationship between adenomyosis and clinical features of abdominal wall endometriosis.*

Characteristic		Adenomyosis (N=26)	No adenomyosis (N=54)	*p*-Value
Age, years	Mean±SD	35.9±5.6	35.8±5.4	0.94
Number of previous cesarean deliveries	Mean±SD	1.62±1	0.98±1.03	0.01
Symptom, n (%)	Dysmenorrhea	18 (69%)	29 (53.7%)	0.14
	Abdominal pain	22 (85%)	48 (89%)	0.7
	Swelling	12 (46%)	19 (35%)	0.24
	Cyclic bleeding from nodule	5 (19%)	9 (16.6%)	0.5
Localization of nodule	Cesarean scar	21 (81%)	32 (59%)	0.12
	Umbilical	5 (19%)	17 (31.5%)	
Fascia involvement, n (%)	Yes	13 (50%)	27 (50%)	0.6
Localization according to fascia, n (%)	Epifascial	15 (58%)	44 (81%)	0.03
	Sub fascial/total*	11 (42%)	10 (19%)	
Histological size of nodule, cm	Mean±SD	3.34±1.4	2.55±1.33	0.016

SD: Standard deviation. *Total infiltration of abdominal wall including rectus muscle, fascia, and subcutaneous tissue.

Table IV. *Association between the body mass index (BMI) and clinical characteristics.*

Characteristic		BMI <30 kg/m^2 (N=59)	BMI ≥30 kg/m^2 (N=17)	*p*-Value
Nodule size, histological	Mean±SD	2.8±1.29	2.74±1.7	0.87
Symptom, n (%)	Abdominal pain	51 (86.4%)	15 (88.2%)	1
	Swelling	22 (37.2%)	6 (35.3%)	1
	Bleeding	11 (18.6%)	3 (17.6%)	1
Localization according to fascia, n (%)	Sub fascial/total*	18 (30.5%)	3 (17.6%)	0.36
Localization of nodule, n (%)	Cesarean scar	36 (61%)	13 (76.4%)	0.44
	Umbilical	18 (30.5%)	4 (23.6%)	
	Other	5 (22.7%)	0	

*Total infiltration of abdominal wall including rectus muscle, fascia, and subcutaneous tissue.

rate from previous reports is 4.5% (15). In our series, only one patient had a recurrence (1.25%). Some authors reported the use of oral progesterone or gonadotropin-releasing hormone analogs, with a low rate of success, and high recurrence of symptoms after cessation of treatment (6). In general, patients with a high level of cell proliferation (Ki67 marker) in endometrial lesions have a significantly higher response rate to hormone therapy than patients with low or moderate proliferation (21). Thus, the proliferation marker Ki67 represents an important predictive marker for the success of hormone therapy and helps in the optimization of endometriosis therapy (21). In our series, in 78% of cases, the proliferation level was less than 30% (Table II). The low rate of success of medical treatment in patients with AWE might be explained by the low proliferation of endometrial tissue of AWE. For this reason, complete surgical excision is the most appropriate therapy in the majority of cases of AWE.

Both ultrasound and MRI are appropriate methods for the assessment of AWE (4). In the present study, both these methods gave accurate results, however, ultrasound is a cost-

Table V. *The proposed classification of abdominal wall endometriosis. Examples of classification: CS b 1: Abdominal wall endometriosis located on the cesarean scar with involvement of rectus muscle, nodule size 2 cm. U1, a 2: Umbilical endometriosis with no previous surgery (primary umbilical endometriosis) epifascial, nodule size 4 cm.*

Factor		Localization				
		CS	T	U1	U2	O
Depth	Epifascial	a	a	a	a	a
	Subfascial	b	b	b	b	b
	Total	c	c	c	c	c
Size*	<3 cm	1	1	1	1	1
	≥3 cm	2	2	2	2	2

CS: Cesarean scar, T: accessory trocar; U: umbilical; U1: pure/primary endometriosis; U2: with previous surgery; O: other localization. *Histological.

effective method. For this reason, ultrasound can be used as a first-line tool for preoperative assessment and MRI can be taken into account in special cases. MRI can be a useful

Table VI. *Overview of the main clinicopathological features related to each localization based on the current study.*

Classification according to localization	Clinicopathological features based on current study
Cesarean scar: CS	Most common type of abdominal wall endometriosis Leading symptom: abdominal pain Coexistence of adenomyosis or pelvic endometriosis in 50% Fascial involvement in 60%
Trocar site: T	Observed at accessory trocar site History of previous surgery (due mainly to endometriosis) Usually on left side
Umbilical: U	Leading symptoms: Cyclic bleeding from nodule, pain Coexistence of adenomyosis or pelvic endometriosis in 82% All epifascial
Primary: U1	No history of previous surgeries (cesarean or laparoscopic surgery for endometriosis)
Secondary: U2	With history of previous surgery
Other: O	Localized on abdominal wall, far from cesarean scar and umbilicus History of previous cesarean section

diagnostic method in small endometriotic nodules that are difficult to palpate, especially in obese women (22). Moreover, MRI can evaluate the relationship between nodules and adjacent structures, which could be helpful for appropriate presurgical planning in cases with possible abdominal wall reconstruction (23, 24). In our suggestion for classification of AWE, the depth of invasion was based on the fascia (Table V). With MRI evaluation, our suggested classification can also be useful in preoperative evaluation, in terms of disease invasion related to the fascia. In contrast to previous reported classifications of AWE related to the depth of disease, including superficial, or fascia type; intermediate, or muscle type; and deep, or peritoneal type (25), in our classification, our definition of "total infiltration" can appropriately describe the invasion of the abdominal wall.

Conclusion

AWE is a rare condition with a high prevalence of symptoms such as abdominal wall pain and swelling, as well as bleeding. The origin and characteristics of AWE are poorly understood. The strengths of the current study are the investigation of the proliferation marker Ki67 in AWE, the finding of the impact of adenomyosis, as well as the suggested classification. Table VI shows the overview of main clinicopathological features based on the current study related to each localization.

Conflicts of Interest

The Authors have no conflicts of interest in this work.

Authors' Contributions

EP, AN and IM: Article writing, data management, data analysis. AM: data management. IM, OG, SS and TR: project development and administration.

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