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Abdominal Wall Endometriosis after Gynaecological Interventions - A Cohort Study on Diagnostic and Treatment of Abdominal Wall Endometriosis

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Abstract

Introduction: Endometriosis is defined as the existence of ectopic endometrial tissue outside the uterine cavity. Endometriosis in the abdominal wall is painful and mainly emerges after surgical abdominal interventions such as laparoscopy, hysterectomy or caesarean sections. The increasing number of surgeries, caesarean section in particular, raises the incidence of abdominal wall endometriosis.

Material and methods: In this retrospective cohort study, the data of women who underwent surgery for abdominal wall endometriosis between 2010 and 2015 has been analysed at the Department of Obstetrics and Gynaecology of the University Medical Centre Freiburg.

Results: Fourteen women were treated for abdominal wall endometriosis during the study period. Median age was 34 (range 27-43). Thirteen women had a previous caesarean section. Lower, cycle-dependent abdominal pain was depicted as the most common reason for consultation. None was diagnosed for endometriosis previously. All women underwent wide excision, and the median tumour weight was 19.9 (range 3-52) g.

Conclusions: Abdominal wall endometriosis is a rare disease. In women with cyclic pain and a history of hysterotomy, abdominal wall endometriosis is a probable cause, and it is easy to cure.

Keywords

Caesarean, Endometriosis, Dysmenorrhea, Ultrasound, Uterine Scar, Endoscopic surgery

Abbreviations

AWE: Abdominal Wall Endometriosis; CT: Computer Tomography; MRI: Magnetic Resonance Imaging

Introduction

Endometriosis is defined as the existence of functional endometrium outside the uterus [1]. Endometriosis lesions are mainly located in the pelvis. However, they can be found almost anywhere in the body. Abdominal wall endometriosis (AWE), as functional endometrium tissue in the abdominal wall, has first been described in the 1950s [2-5]. Endometriosis lesions in the front abdominal wall are rare and are reported after abdominal interventions such as laparoscopy, caesarean section, tubal ligation, hysterectomy and amniocentesis [6-11]. The prevalence of AWE in women with preceding gynaecologic interventions is 0.03-1.08% [12,13] and therefore only depicts a small part of extra pelvic endometriosis [9]. Nominato et al. consider caesarean sections as a main risk factor for the occurrence of AWE [12]. Because symptoms are heterogeneous and imaging can be difficult to interpret, clinical diagnosis can be challenging [14,15]. Etiopathogenesis of endometriosis is still a matter of debate. AWE, however, involves a direct transplantation of the functional endometriosis as the most probable mechanism for the genesis after an abdominal intervention with uterus opening (secondary AWE) [16]. There were also cases described without any preceding surgical intervention (primary AWE).

A subcutaneously located AWE lesion is palpable through the abdominal wall. Diagnosis is mainly suspected when the pain is cyclic. The lesions might be visible as a brown, blue, violet or even black subcutaneous spot. Additional medical imaging could be used to verify the suspected diagnosis of AWE. In sonography, subcutaneous endometriosis lesions present themselves as little echoic and/or solid, and sometimes cystic tumours [15,17]. The margins are bounded in a blurry way, not relocatable and often seem to infiltrate the adjacent tissue [18,19]. Infiltration of neighbouring structures or a distinction from differential diagnoses could be evaluable via computer tomography (CT) [20] or magnetic resonance imaging (MRI). Possible differential diagnoses are granuloma, hematoma, lymphoma or lipoma.

Finally, the diagnosis is confirmed through histopathology. Local wide excision is the gold standard of therapy. A total resection with clear margins should be achieved.

Further, there is a recurrence of 4% of cases. A conservative therapy including GnRH analogues, combined oral contraceptives or progestin-only pill, can reduce symptoms, but cessation of medication leads to recurrence of symptoms. Surgery, in case of clear margins, is the unavoidable treatment of choice.

Materials and Methods

A retrospective analysis of women treated for a histologically

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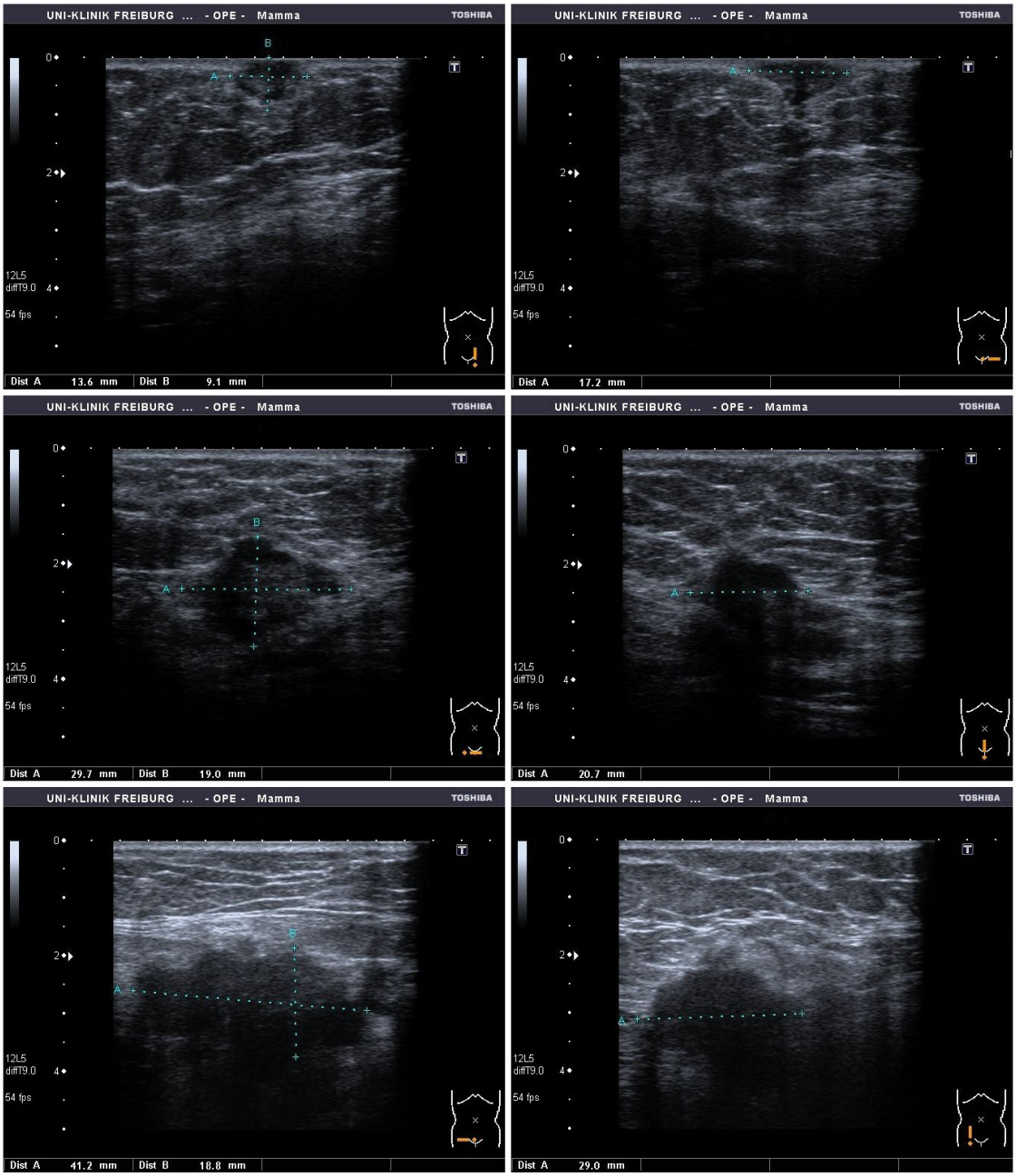

Figure 1: Ultrasonography (2D, Aplio 500, 13 MHz (Toshiba™)): 3 cases of AWE in the subcutaneous tissue layer at Pfannenstiel incision after caesarean section. Top: AWE in the subcutis, middle and bottom: AWE in the fascia of the M. rectus abdominis.

Table 1: Characteristics of the patients with AWE.

	Mean (Range)
Age, years	33.6 (27-43)
BMI, kg/m²	25.6 (18.9-33.9)
	n
Prior vaginal birth/VE	4
Prior caesarean section	13

confirmed AWE has been conducted at the Department of Obstetrics and Gynaecology of the University Medical Centre Freiburg between 2010 and 2015. Data on their age, symptoms, preceding surgeries, deliveries, previously diagnosed or treated endometriosis preoperative diagnostics, localisation and size of the endometriosis, lesion, histology, infiltration of neighbouring structures, operation time, operational approach and follow-up were included. Descriptive data ascertainment and one-dimensional frequency distribution were conducted.

The study was approved by the ethical committee of Freiburg University, registration number 35/16.

Results

The analysis included 14 women and their clinic features are summarised in table 1. The average age was 34 years (27-43 years). Moreover, 13 out of 14 women had at least one Pfannenstiel incision for caesarean section in their anamnesis. None of the women had been treated for pelvic endometriosis or had an adenomyosis uteri operation previously. Two women, however, underwent surgery because of a recurrent AWE.

The most common symptom of AWE was pain, which was described by all women (n = 8, no data (N/D) = 6)). Seven out of eight women suffered from cyclic pain. AWE lesion was palpable in all patients. Table 2 summarises the symptoms. One woman first presented at the general surgery department with a suspected laparoscopic trocar port-site hernia. In the remaining cases, the first presentation was at a gynaecologist.

Abdominal 2D ultrasonography (US) on every patient (Figure 1) and an additional magnetic resonance imaging (MRI) of the pelvis in one patient (Figure 2) were conducted before surgery.

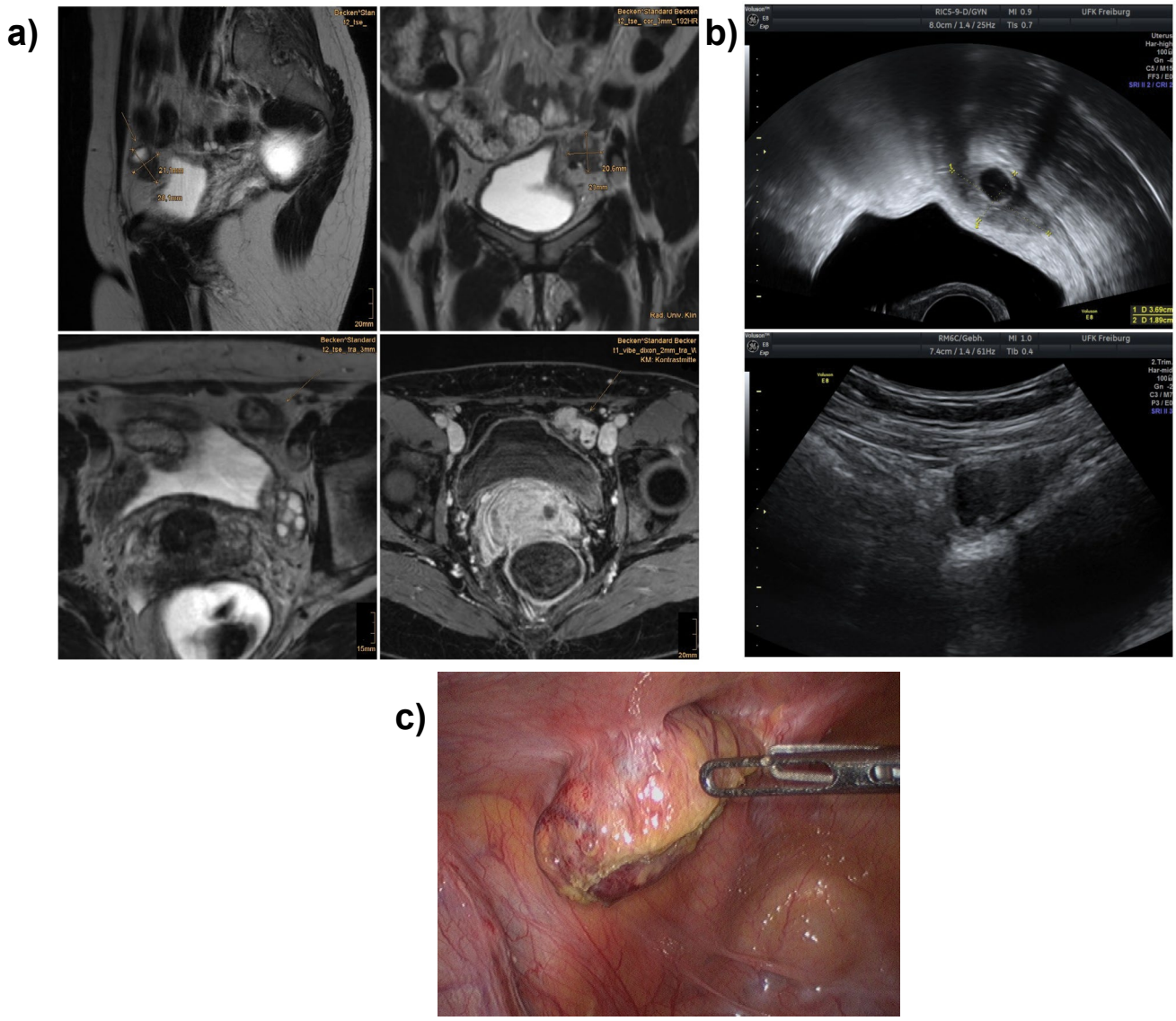

Figure 2: Case of a 43-year old patient with AWE left lateral of the bladder after laparoscopic supracervical hysterectomy (LASH) and cervicopexy.

a) MRI of the pelvis, 1x multihance 15 ml (MU15): Endometrioma with a myomatous component left lateral of the bladder at the parietal peritoneum, 3.1x 3.0 cm, cystic. T1 hyperintense parts manifest contrast agent enhancement; b) Top: transvaginal ultrasonography (2D, Voluson E8 GE healthcare™, 5-9 Mhz); bottom: transabdominal ultrasonography (2D, Voluson E8 GE ealthcare™); c) Intraoperative presentation of the endometrioma.

Table 2: Clinical characteristics of the patients with AWE.

Presenting symptoms	N (%)	No data (n)
Palpable mass	10 (91%)	3
Pain	8 (100%)	6
Pain with cyclicity	8 (89%)	5
(Livid) Discoloration	1 (14%)	7

Table 3: Histopathological characteristics of the excised AWE specimens.

Characteristics of the excised AWE	Mean (Range)
Number	1.36 (1-3)
Weight, g	19.9 (3-52)
Volume, cm^3	5.1 (0.3-13.6)

Before surgery, AWE was suspected to be the cause for complaint all cases. All surgeries were performed with laryngeal mask as general anaesthesia. The average operation time was 38 minutes (11-126 minutes). In 13 out of 14 cases, the lesion was removed by preparation along the wire marking. In one case, the resection was performed laparoscopically. Before surgery, the lesions were marked with a wire under sonographic control to facilitate intraoperative identification of the tumour (Figure 3 and Figure 4).

The histological assessment of the resected lesions is illustrated in table 3. A clinical and histological infiltration of the endometriosis in the frontal and/or side abdominal wall musculature (M. rectus abdominis or M. obliquus externus/internus) or its aponeurosis was described in 43% of the women. Figure 5 shows an AWE specimen.

One of the patients had a hormonal treatment (dienogest) after surgery and demonstrated no recurrence during follow-up. The average follow-up time of all patients was 20 months (1-59). None of the patients was diagnosed with pelvic endometriosis during follow-up. Two women reported a recurrence of complaints in the resected area of the AWE. The complaints emerged about 6 months after surgery. Both patients had not received any hormonal treatment after surgery. The case reported in illustration 5 demonstrates the possible steps in diagnosing AWE in a postoperative scar.

Reimplantation did not apply because they did not have any abdominal surgical interventions between the first and second AWE.

Discussion

Endometriosis of the abdominal wall is rare and mostly emerges after surgical intervention along the abdominal incision scar site in the subcutaneous tissue layer [6-11]. Postoperative AWE emerges after various surgical interventions, most often after a caesarean section. The prevalence of AWE after gynaecological surgery is stated as 0.03-1.08% [12,13], and, therefore, it only illustrates a small part of extra pelvic endometriosis. However, there are cases of AWE without any preceding surgical intervention [11].

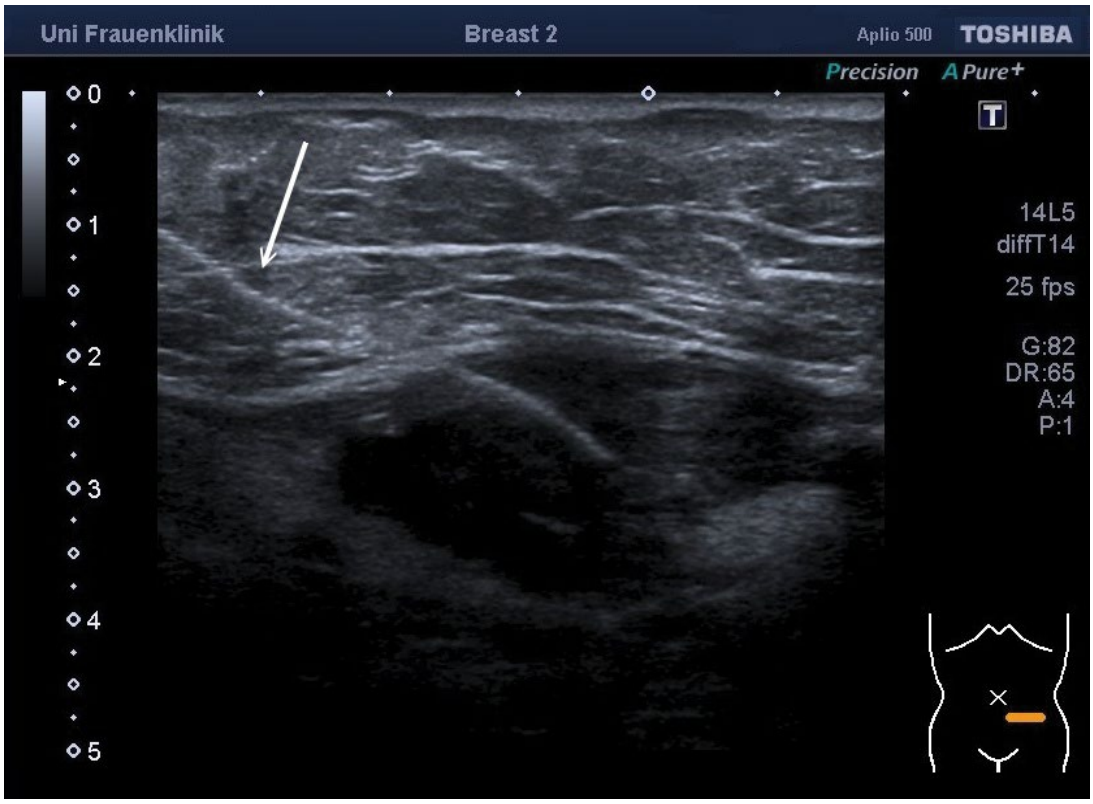

Figure 3: Preoperative ultrasound-guided wire marking (2D, Aplio 500, 13 MHz (Toshiba™)).

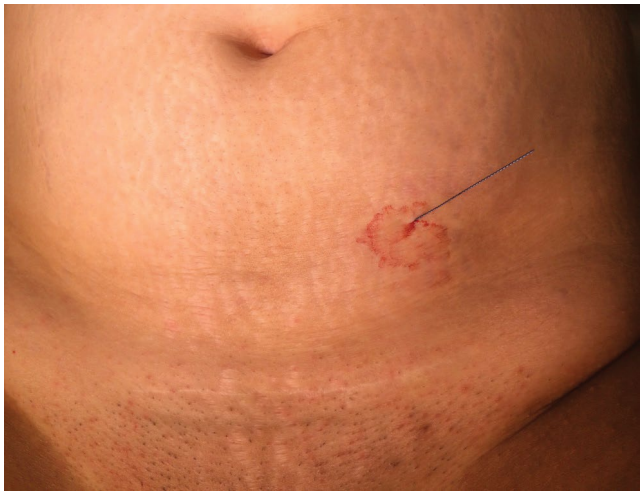

Figure 4: Preoperative finding of the wire marked AWE lesion. The AWE lesion is situated in the lateral edge of the fascial incision, not in the cutaneous scar.

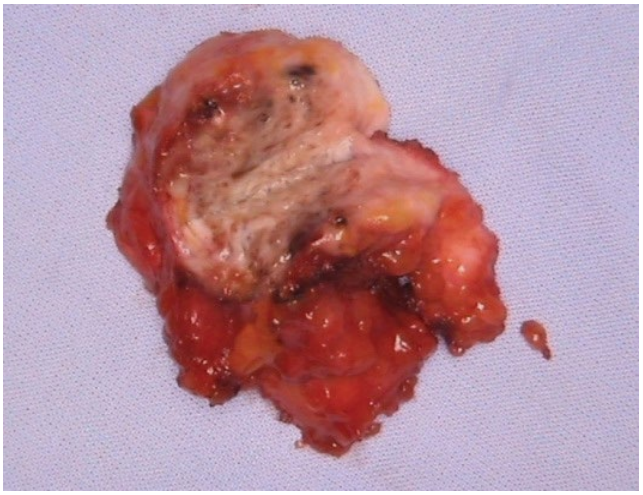

Figure 5: AWE specimen (4x3.5x2 cm).

Endometriosis is associated with increased risk of various malignancies with the best evidence for ovarian cancer. The most common histopathological subtype is clear cell cancer (CCC) or endometrioid cancer (EC) [21]. The risk of malignancy in AWE is described as 0.31% [22]. Malignant transformation in the reported cases occurred between three and 39 years after endometriosis had been diagnosed [23]. Taburiaux performed a review on endometriosis-associated abdominal wall cancer in 2015 [24]. Most common histological subtypes were CCC in 63% and EC in 22% of patients. Da Ines et al. reported on a mixed endometrioid and serous carcinoma arising from AWE after caesarean section in a 48-year old 16 and 20 years after caesarean section. A whole-body positron emission tomography showed suspicious left iliac nodes. Histopathology of the lymph nodes revealed subcapsular micro-metastasis, leading to a multidisciplinary recommendation of adjuvant chemotherapy [25]. Patients presented with a fast growing tumour, sometimes recurring and resistant against medical treatment [26]. Radical surgery to obtain healthy margins is considered the gold standard [26]. Chemotherapy with carboplatin and paclitaxel has been most frequently described. Radiotherapy or progestin therapy with various treatment protocols have been applied [24,26-28]. The prognosis seems to be poor as the median survival time after diagnosis was 30 months [24].

Endometriosis can also present in the scar of an episiotomy after a vaginal delivery. It is likely that viable endometrial cells are mechanically transplanted into the episiotomy wound during vaginal delivery. As in AWE, patients present with a palpable painful lesion. Local wide excision is important to prevent recurrence. Li et al. state that hormonal suppression after surgery seems not to be effective to prevent recurrence [29].

The etiopathogenesis of endometriosis is still unclear and controversial. Primarily, the proposed hypotheses are based on two models [30]: the implantation theory and the metaplasia theory. The first one is considered as a secondary implantation (after retrograde

menstruation or iatrogenic direct implantation) of endometrial cells into extrauterine tissue. During hysterotomy endometrial cells can inoculate to the peritoneum or the abdominal wall [31]. The second describes a spreading of cells in the early embryonic stage and the movement of those along the coelomic cavity.

Although there is no history of pelvic endometriosis in AWE patients, the iatrogenic implantation seems the most probable mechanism for scar endometriosis after abdominal interventions [16]. In this retrospective analysis, all patients had previous surgery, which was a likely origin of AWE. None of our patients was diagnosed with endometriosis earlier or presented with symptoms referring to pelvic endometriosis during follow-up. Two patients suffered from cycle-dependent complaints within the scar region within 6 months after AWE removal. This finding highlights the relevance of a complete resection of the AWE. The incidence of pelvic endometriosis is similar in women with and without AWE and is in the order of 8-15% [32,33]. As none of our patients had a diagnostic laparoscopy to confirm or confound pelvic endometriosis before or during follow-up, the relation between pelvic and AWE is uncertain. It is hypothesised that endometrial tissue protracted from the uterine cavity after hysterotomy might preferentially implant in injured but sufficiently vasculated tissue such as the abdominal wall muscle but not in the sound peritoneum of the pelvis. Another hypothesis is that, if hysterotomy leads to a spread of endometrial cells into the pelvis and the abdominal wall, endometriomas in the abdominal wall become painful earlier. Further studies with laparoscopic inspection are therefore warranted.

Endometriosis remains a disease that is mostly diagnosed after a long period of suffering, thus making anamnesis ascertainment essential. AWE is a largely clinical diagnosis [31]. A pain journal can render assistant as cycle-dependent pains in the surgical scar region can lead to a diagnosis. Women without prior caesarean section often have AWE lesions at the umbilicus, whereas women with a prior caesarean section are more likely to have AWE in the incisional right (36, 5%), left (46, 2%) or midline (11, 5%) [31]. AWE often presents in the edge of the suture of the abdominal muscle fascia that is far more lateral than the edge of the scar in the skin. Hence, AWE and the underlying surgical intervention as reason for the complaints is not considered because the pain is not in the area of the scar seen on the abdomen (see illustration 4). AWE is rare and often not considered as a cause of abdominal pain, and symptoms are inconsistent, making its prevalence difficult to quantify. In our patients, pain was the most frequent symptom occurring cyclically. AWE was palpable in all of the patients. A multifrequency linear transducer (7.0-13.0 MHz), which is also used for breast sonography, is more suitable for the assessment of an AWE nodule in the fascia of the M. rectus abdominis region than the transabdominal convex probe. Ultrasound and a detailed anamnesis are sufficient to diagnose AWE. In ultrasound AWE presents as a hypoechoic nodule with speculated margins infiltrating the surrounding tissue [34]. At colour Doppler examination a single vascular pedicle often enters the nodule, abundant intralesional vascularization can be seen [19]. CT and MRI can be additionally used for the diagnosis [35]. Endometriosis can be difficult to diagnose via CT or MRI [15], and misinterpretation of results can lead to misdiagnosis, over-diagnosis and overtreatment. Lack of expertise in the diagnosing tools could lead to the suspicion of a malignancy, with the operation being performed too extensively, which could be stress- or harmful for the patient. During sonography a fine-needle aspiration for cytology can be performed, but is inconclusive in up to 75% of the cases [31]. Preoperative ultrasound-guided wire marking can facilitate intraoperative identification of the lesion. Granulomas, abscesses, hernias, lipomas, atheroma, dermoid and malign tumours such as soft-tissue sarcomas or subcutaneous metastases need to be distinguished as a differential diagnosis. The chosen therapy depicts the vast local excision with sufficient safe distance to prevent recurrences. In the case of widespread resections of the fascia, a repair using mesh might be indicated [36]. However, this was not necessary in our patients. There were no abdominal wall malfunctions after surgery. Medical therapy can be an addition but never an alternative to surgery because symptoms reoccur after discontinuation [36]. Optional, hormonal treatment before surgery may diminish the endometrium and reduce the pain before surgery. Darwish et al. report their experience with triptorelin, a gonadotrophin releasing hormone agonist in combination with add-back therapy by percutaneous estradiol for treating endometriosis at the episiotomy site [37]. In this case report the nodule was too close to the anal sphincter to be resected with clear margins at the time of diagnosis. Nodule size and the therewith-associated complaints reduced significantly under the treatment, which will be proceeded until menopause. The authors submit the disadvantage of high expenses of this therapy. Concerning medical management of abdominal wall endometriosis Koger et al. reported a series of 11 patients who failed oral progestogen therapy, but had complete resolution of symptoms after surgical excision [38]. Chatterjee reported on another series of 11 patients that failed to respond to a 4- to 6-month course of norethisterone. All 11 were treated successfully with surgery [13]. To date, there are no data to support postoperative hormonal therapy. However, this may be appropriate in patients with a history consistent with pelvic endometriosis [11].

Yet there are no guidelines for the prophylaxis of AWE after gynaecological surgeries. The extensive peritoneal lavage after uterine closure, suture of the parietal peritoneum, the change of surgical instruments, uterine closure or the covering of the edges of the fascia of the abdominal muscle before hysterotomy are discussed in the literature, as is the role of the externalisation of the uterus during surgery. None of these approaches has been prospectively examined in clinical trials [11,39].

In summary, AWE as causative factor for pain in women after gynaecological surgeries must be considered, even when the localisation of the complaint is not at the cutaneous scar (Figure 4). AWE is largely a clinical diagnosis. Complete local excision is the recommended treatment of choice to achieve no recurrences of symptoms in the long-term. Gynaecologists should take into account the increasing incidence of the AWE, focusing on its diagnosis and appropriate treatment.

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