

Bifocal endometriomas involving a Pfannenstiel incision. Clinical case

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Endometrioma of the anterior abdominal wall (EAAW) is a rather rare variant of extragenital endometriosis, which in most cases occurs after obstetrical and gynecological procedures. EAAW presented predominantly as a single tumour-like mass, and multiple ectopias were observed in only 1.9–5.6% of cases, exclusively after Pfannenstiel laparotomy.

Here we present a clinical case of a 37-year-old patient who complained of the large tumour-like nodules along the postoperative anterior abdominal wall scar, accompanied by severe cyclic, catamenial pain. Additionally, the patient noted an increase in tumour size during menstruation. Thirty-three months ago, she underwent an elected cesarean section for obstetric indications. Based on ultrasonography and computed tomography scans, the presence of two EAAW in the corners of the postoperative scar was established: 46 × 32 × 31 mm and 14 × 18 × 13 mm, respectively. Both lesions were excised out without damaging their integrity. The fascial defect was replaced by synthetic polypropylene mesh. The diagnosis of EAAW was finally confirmed based on pathological (presence of endometrial glands and cytotogenic stroma) and immunohistochemical (positive membrane expression of CD10 in cytotogenic stroma, intense cytoplasmic expression of CK7 in endometrial glands, marked nuclear expression of progesterone (PR) and estrogen (ER- α) receptors in endometrial glands and cytotogenic stroma, proliferative activity index Ki-67 – 2%) studies. At a follow-up after 19 months, the patient was asymptomatic; according to physical examination and ultrasound scan, there was no evidence of recurrence.

Abdominal wall endometriosis is a rare condition. Clinicians should be aware of this pathology, especially in women presenting with a painful mass near the scar of a previous obstetrical and gynecological surgery. Surgery is the best treatment modality for endometrioma, whereas its optimal volume is considered to be R0 resection with preservation of endometriomas' integrity. The final diagnosis of EAAW requires pathological and immunohistochemical confirmation.

KEYWORDS

extrapelvic endometriosis, scar endometriosis, abdominal wall endometriosis, caesarean section.

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Endometrioma of the anterior abdominal wall (EAAW) is a rather rare and specific variant of extragenital endometriosis [6, 12], with a frequency of 0.4% to 3% in the structure of endometriosis in women of reproductive age [3, 12]. In most cases, EAAW occurs after obstetric and gynecologic surgical interventions (cesarean section, myomectomy, hysterectomy, laparoscopy) [2, 6, 7, 12–14]; however, cases of condition after general surgical procedures also have been

described [3, 11], as well as sporadic observations of spontaneous EAAW have been published [3, 5, 6, 9, 10].

EAAW presented predominantly as a single tumour-like mass, and multiple ectopias were observed in only 1.9–5.6% of cases, exclusively after Pfannenstiel laparotomy [4, 15]. Due to the rarity of multiple EAAW, we present our own clinical observation with emphasis on the diagnosis and surgical treatment of this condition.

Case report

A 37-year-old patient (G3P3) complained of tumour-like masses in the area of the anterior abdominal wall postoperative scar, accompanied by marked cyclic, catamenial pain. Additionally, the patient noted an increase in tumour-like masses during menstruation. The length of the disease was 12 months when the above-described phenomena first appeared. Menarche at 13 years, menstrual cycle of 28 days, menstruation of 4–5 days, painless. Thirty-three months ago, she had an elected cesarean section for obstetric reasons. Two previous pregnancies ended in timely deliveries through natural labour.

On examination, the patient has Class II obesity (BMI – 36.05 kg/m²). In the area of the left corner of the Pfannenstiel incision, a moderately mobile and slightly painful tumour-like mass approximately 5 cm in size is detected. On the right end of Pfannenstiel's incision, a second tumour-like mass about 2 cm in diameter, mobile and somewhat painful, is determined. The skin over both masses was unchanged.

On ultrasonography (USG) in the left iliac region, directly under the postoperative scar, a solid (38.1 mm) hypogenic mass of oval shape with a cellular texture and a clear hyperechogenic contour was detected. In Doppler mapping, peripheral and central blood flow (degree of vascularization – 2) and the presence of an afferent vessel with a diameter of 3 mm are determined in the mass. On the right side, a hypogenic mass (26.4 mm) with a hyperechogenic contour was visualized in the subcutaneous tissue of the anterior abdominal wall; unexpressed peripheral blood flow was detected at Dopplerography.

A computed tomography (CT) scan was performed on the second day after menstruation to determine the prevalence of tumour-like masses of the

anterior abdominal wall. In the right iliac region, a solid mass measuring 14 × 18 × 13 mm is located in the subcutaneous tissue; on the left, a solid mass (46 × 32 × 31 mm) with a clear and irregular contour involving subcutaneous tissue, fascia, and left rectus abdominis muscle (Fig. 1). A preoperative diagnosis of multiple endometriomas of the anterior abdominal wall after cesarean section was established.

The surgical intervention was planned and performed in the middle of the patient's menstrual cycle. Revision on the left side revealed a tumour-like mass with involvement of subcutaneous tissue, fascia and rectus abdominal muscle. Excision of the tumour-like nodule en bloc within healthy tissues was performed; the fascial defect was replaced with polypropylene mesh. The tumour-like mass on the right was excised within the subcutaneous tissue. Both anterior abdominal wall masses were removed without compromising their integrity. The surgery was completed with layer-by-layer suturing of the anterior abdominal wall tissues.

In section, the gross specimens are represented by multiple microcavities with hemorrhagic contents (Fig. 2). Microscopic examination of both masses noted the presence of endometrial glands and cytogenic stroma (Fig. 3).

Immunohistochemical study (Fig. 4, 5) revealed positive membrane expression of CD10 in cytogenic stroma, intense cytoplasmic expression of CK7 in endometrial glands, marked nuclear expression of progesterone (PR) and estrogen (ER-α) receptors in endometrial glands and cytogenic stroma, and proliferative activity index (Ki-67) – 2%. Thus, pathomorphologic findings fully confirmed the diagnosis of anterior abdominal wall endometriomas.

The postoperative period was uneventful, and the patient was discharged from the hospital on the third postoperative day. At the follow-up

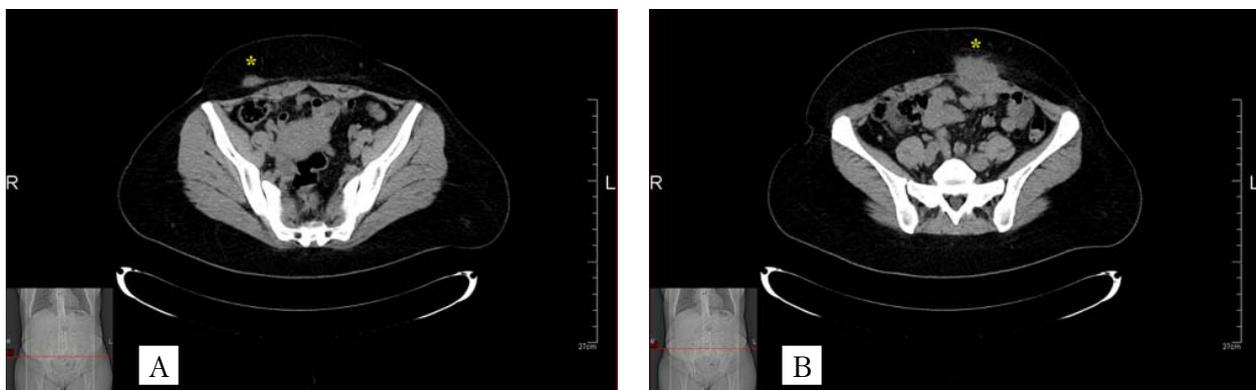


Figure 1. Axial computed tomography: solid mass in the subcutaneous tissue of the right iliac region (A); solid mass of the left iliac region, involving the subcutaneous tissue, fascia, and left rectus abdominis muscle (B)

* Solid mass (endometrioma)



Figure 2. **Macroscopic appearance of abdominal wall endometriomas**

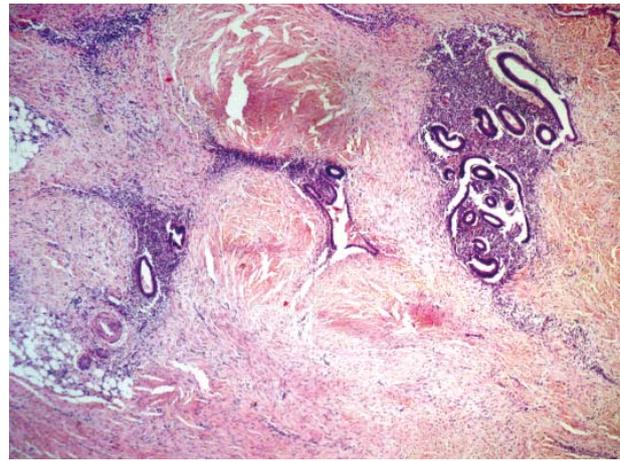


Figure 3. **Microscopic examination: endometrial glands and cytotrogenic stroma.** H&E staining, $\times 40$

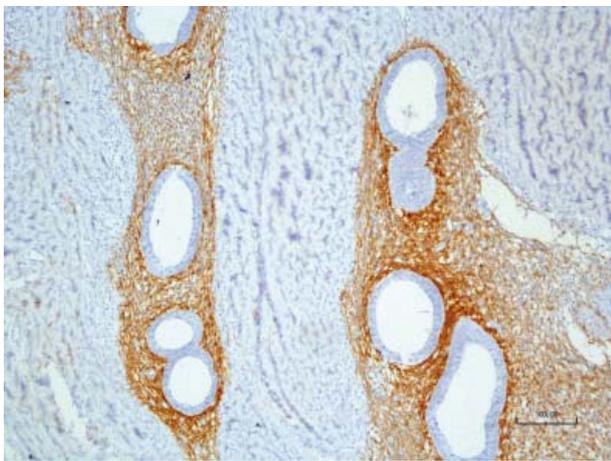


Figure 4. **Immunohistochemistry: diffuse CD10 expression in cytotrogenic stroma.** DAB $\times 100$



Figure 5. **Immunohistochemistry: diffuse expression of PR in endometrial glands and cytotrogenic stroma.** DAB $\times 40$

examination after 19 months, the patient was asymptomatic, and according to the results of the physical examination and ultrasound scan, there was no evidence of recurrence.

Discussion

Since the first description of EAAW by Meyer R. in 1903, less than 1000 cases have been published in the specialized literature [11]. The age of patients with EAAW ranged from 21 to 49 years [2–4, 7, 8, 11, 12, 14, 15], and the average time from the primary surgical intervention to diagnosis of EAAW was 28.3–38.6 months [1, 6, 15], although in a number of studies this index varied quite widely, from 5 to 20 years [2, 3, 8, 9, 14].

The etiopathogenesis of EAAW has not been definitively studied to date [1, 6, 13]. However, autotransplantation of endometrium during surgical interventions on pelvic organs is believed to be the

most probable trigger in the occurrence of EAAW – the so-called implantation theory [2, 4, 8]. At the same time, the assumption of possible hematogenous and lymphatic spread, as well as the concept of metaplasia of primitive polypotential mesenchymal cells, are the basis for justifying the occurrence of spontaneous EAAW [1, 4, 6].

It has been noted that one of the potential factors associated with EAAW is increased body weight (BMI ≥ 25 kg/m²) in operated women [2, 3, 13], which is usually explained by additional technical difficulties in performing surgical interventions in this category of patients, namely, inadequate hysterorrhaphy and tissue contamination of the anterior abdominal wall [2].

The vast majority of cases (from 59% to 100%) of EAAW were observed after cesarean section [2, 3, 6–9, 11–15], and in view of the global growth of cesarean section, an increase in this pathology should be expected [1, 8]. However, the current

incidence of EAAW after cesarean section is quite low and is estimated to be between 0.21–1.3% [1, 8], which presumably should be explained by the potentially large number of undiagnosed cases [2].

To date, there are no evidence-based studies on the prevention of EAAW after cesarean section [1], but some publications recommend the following measures: (1) use of wound protectors/retractors; (2) use of separate suture materials when suturing the uterus and the anterior abdominal wall; (3) avoiding contact of gauze swabs used inside the uterine cavity with the soft tissues of anterior abdominal wall, or even complete renouncement from their use; (4) changing gloves and surgical instruments after suturing the uterine wall; and (5) performing thorough cleaning of subcutaneous tissue and fascia, especially at the corners of the Pfannenstiel wound, at the final stage of surgery [2, 7, 15].

Classically, EAAW is characterized by a symptomatic triad: catamenial pain, palpable mass within the anterior abdominal wall, and history of obstetric-gynecological interventions [1, 2, 6, 7]. However, these signs may not always be identified. Thus, cyclic (catamenial) pain is observed in 51.0–86.9% of EAAW cases [2, 3, 7, 8, 11–13, 15], and an anterior abdominal wall mass is recognized on palpation in only 32.5–66.7% of observations, which depends on depth of its location within the anterior abdominal wall, i.e., in subcutaneous tissue, fascia, rectus abdominis muscle, or peritoneum [2, 3, 7]. External catamenial bleeding from EAAW has been described as a rare symptom (1.25–9.1%) [11, 13]. As a result of the low specificity of symptoms, the diagnosis of endometrioma is often difficult, and initially EAAW are treated as hernias, lipomas, granulomas, desmoid tumours, etc. [1, 3, 4, 7, 9, 11].

When EAAW is suspected before surgery, it is advisable to use medical imaging studies in order to clarify the size and spread of mass within the anterior abdominal wall tissues and, if aponeurosis is involved, to anticipate the necessity of replacing the fascial defect using synthetic meshes [3, 6]. Currently, USG is considered the first-line diagnostic method of EAAW, and its informativity is 78.3–97.0% [1–3, 6, 11–]. EAAW has been visualized as a hypogenic mass with a hyperechoic contour and varying degree of vascularization from the periphery toward the centre according to Doppler USG [2, 6].

Magnetic resonance imaging (MRI) and CT are preferred tools for diagnosis and planning the extent of surgical procedure for deep and widespread EAAW, as well as for nonpalpable masses of the anterior abdominal wall [1–3, 6, 11–14]. EAAW, according to MRI on T1W (with and without fat suppression) and T2W images, is visualized

as a hyperintense heterogeneous mass in the area of the postoperative scar as a result of catamenial hemorrhages in endometrial ectopia. With prolonged conservation of EAAW, its radiological characteristics change due to progressive hemosiderin deposition and pronounced fibrosis; this determines low signal intensity on T2W images [1, 4]. On CT, EAAW is visualized as a solid mass in the area of the postoperative anterior abdominal wall scar [2, 9]. Possible radiologic appearance depends on the menstrual cycle phase, ratio of stromal and glandular elements, amount of hemorrhagic imbibition of ectopia, and degree of inflammation and fibrosis in the surrounding tissues [9].

In most cases, EAAW is located directly in the thickness of the postoperative scar after Pfannenstiel laparotomy, predominantly in the left corner (66%), and in rare cases, ectopias at a 5–6 cm away from the former incision line have been described [3, 13]. According to the localization in layers of the anterior abdominal wall, EAAW are divided into: superficial – placed in subcutaneous tissue (above the aponeurosis); intermediate – with infiltration of fascia and rectus abdominal muscle sheet; and deep – when ectopia is located in the thickness of the rectus abdominis or involved peritoneum [2]. As maintained by E. Piriye et al., out of 80 cases of EAAW, ectopias were localized predominantly epifascial (72.5%) and in 27.5% – subfascial or with involvement of all anterior abdominal wall layers [13]. According to a number of studies, involvement of fascia in endometrial ectopia was noted in 60.0–71.4% of EAAW cases [3, 13, 15].

EAAWs are mostly located in the corners of the postoperative scar after Pfannenstiel laparotomy [11]. A similar pattern was noted in the study of P. Zhang et al. (2019), where localization of endometriomas in the ends of scars after Pfannenstiel laparotomy was observed in 83% of cases and after midline laparotomy – in 84.2%. Moreover, the authors suggested that Pfannenstiel laparotomy is more predisposed to EAAW formation compared to midline laparotomy [15]. The mean size of EAAW in published clinical series was 22–42 mm [2, 3, 7–9, 11, 13], and in few reports, the size of endometrial ectopia reached 14 cm [4].

Fine-needle aspiration biopsy is used by some authors to clarify the diagnosis of EAAW and to exclude malignancy [3, 6, 8, 11, 14]. However, the use of this method for diagnosis of EAAW is rather controversial due to potential implantation along the puncture channel, and therefore, its excision is considered mandatory during subsequent surgery [1, 4, 6].

Actually, the use of hormonal drugs is not considered an independent method for EAAW treatment

as the severity of symptoms decreases only while taking the drugs and recovers after discontinuation of treatment [1, 3, 6]. According to current recommendations, hormones are used in patients who refuse surgery, as well as in premenopausal women or within the postoperative period to prevent recurrence [6, 11].

Surgical excision is the mainstay of treatment for symptomatic EAAW [1, 2, 4, 6]. The generally recognized principles of surgery for EAAW include excision of endometriomas together with surrounding fibrosis within healthy tissue (R0 resection) and thorough preservation of mass integrity to prevent re-implantation of endometrial cells [2, 6]. Therefore, a number of studies recommend maintaining a distance of 5–10 mm from the edges of endometrioma [1, 8].

In cases of aponeurosis excision during surgical removal of EAAW, further approach depends on size of the formed fascial defect. Thus, small defects are sutured according to the tension free principles [1, 2, 6], and polypropylene meshes are recommended to replace aponeurosis defects larger than 3 cm [1, 2, 6, 14]. The frequency of anterior abdominal wall reconstruction using synthetic grafts in different series of patients with EAAW ranged from 6.7% to 36.4% of cases [2, 3, 6–8, 11].

In cases where EAAW is located in the rectus abdominis with spread to the peritoneum, the use of laparoscopic techniques for surgical ablation of endometrial ectopias in the anterior abdominal wall and pelvic peritoneum is recommended [2, 3, 6]. It should be noted that concomitant pelvic endometriosis (adenomyosis) in patients with EAAW can be diagnosed in 8.7–96.2% of cases [3, 9, 12, 13].

Various percutaneous ablation options have been proposed as an alternative to surgical treatment of EAAW, including: (1) USG-controlled sclerotherapy with alcohol, polidocanol; (2) Chemical cauterization with silver nitrate; (3) Laser vaporization; (4) Cryoablation; and (5) High-intensity focused ultrasound [1, 5]. The common disadvantage of the mentioned methods is the inability to confirm diagnosis pathomorphologically and to exclude malignancy [11].

Histopathologic examination of the removed anterior abdominal wall masses is considered to be the final stage of EAAW diagnosis and includes detection of endometrial glands surrounded by cytogenic stroma with foci of hemorrhages (hemosiderin) and characteristic signs of hormonal transformation in the secretory or proliferative phases [4, 7, 9, 11]. Immunohistochemistry with positive expression in cytogenic stroma (CD10, PR, ER α) and endometrial glandular structures (CK7, PR, ER α , Ki67) is

a desirable additional method of EAAW diagnosis [4, 7, 13].

Malignant transformation of EAAWs is a rare phenomenon, with an incidence of less than 1%, and its main histopathological variants are represented by clear cell carcinoma and endometrioid adenocarcinoma [7, 10, 14]. G. Liu et al. (2021), based on a systematic review of the literature, stated that there are currently no standardized treatment protocols for malignant EAAW [10].

Although no recurrence of EAAW has been observed in most studies [2, 4, 8], however, few publications indicate that its incidence can be as high as 3.3–15.4% at follow-up times ranging from 12 to 36 months [7, 11, 12].

Conclusions

Thus, the presence of a tumour-like mass in the postoperative scar after obstetric and gynecological interventions, accompanied by catamenial pain and its increase in volume during menstruation, is characteristic for EAAW. Surgery remains the leading method for EAAW treatment, and its optimal volume is considered R0 resection with preservation of endometrioma integrity. The final diagnosis of EAAW requires pathomorphologic and immunohistochemical confirmation.

DECLARATION OF INTERESTS

The authors declare that they have no conflicts of interest and that they have no financial ties to disclose.

ETHICS APPROVAL AND WRITTEN INFORMED CONSENTS STATEMENTS

Oral and written informed consent was obtained from the patient to publish the patient-related data in anonymized form.

AUTHORS CONTRIBUTIONS

A.E. Mishina: project development, management, manuscript writing; E.V. Gutu: data collection, manuscript editing; I.V. Mishin: project development, data collection, manuscript editing; and S.E. Gutu: data collection, manuscript editing. All authors read and approved the final manuscript.

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Біфокальні ендометріоми з розрізом Пфанненштиля. Клінічний випадок

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Ендометріома передньої черевної стінки (ЕПЧС) — досить рідкісний варіант екстрагенітального ендометріозу, який у більшості випадків виникає після акушерсько-гінекологічних маніпуляцій. ЕПЧС представлений переважно як одна пухлиноподібна маса, а множинні ектопії спостерігалися лише в 1,9—5,6% випадків, виключно після лапаротомії за Пфанненштилем.

Представлено клінічний випадок пацієнтки віком 37 років, яка скаржилася на великі пухлиноподібні вузли вздовж післяопераційного рубця на передній черевній стінці, що супроводжуються сильним циклічним катаменіальним болем. Крім того, пацієнтка відзначила збільшення розмірів пухлини під час менструації. Тридцять три місяці тому їй зробили плановий кесарів розтин за акушерськими показаннями. За даними ультразвукового дослідження та комп'ютерної томографії встановлено наявність у кутах післяопераційного рубця двох ендометріом передньої черевної стінки розмірами 46 × 32 × 31 та 14 × 18 × 13 мм. Обидва вогнища були вирізані без порушення їхньої цілісності. Фасціальний дефект замінено синтетичною поліпропіленовою сіткою. Діагноз ЕПЧС був остаточно підтверджений на основі патологічних (наявність ендометріальних залоз і цитогенної стромі) та імуногістохімічних (позитивна мембранна експресія CD10 в цитогенній стромі, інтенсивна цитоплазматична експресія СК7 в ендометріальних залозах, помітна ядерна експресія прогестерону (PR) і естрогену (ER-α) рецепторів в ендометріальних залозах і цитогенній стромі, дослідження індексу проліферативної активності Ki-67 — 2%). Під час контрольного обстеження через 19 міс пацієнтка була безсимптомною. За даними фізикального обстеження та ультразвукового сканування не виявлено жодних ознак рецидиву.

Ендометріоз черевної стінки є рідкісним захворюванням. Клініцисти повинні знати про цю патологію, особливо у жінок з хворобливою пухлиною біля рубця після попередньої акушерської та гінекологічної операції. Найкращим методом лікування ендометріоми є хірургічне втручання, оптимальним його обсягом вважають резекцію R0 зі збереженням цілісності ендометріоми. Остаточний діагноз ЕПЧС потребує патологічного та імуногістохімічного підтвердження.

Ключові слова: позатазовий ендометріоз, рубцевий ендометріоз, ендометріоз черевної стінки, кесарів розтин.

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