PRACE KAZUISTYCZNE
położnictwo

Giant recurrent perineal endometriosis in an episiotomy scar – a case report

Olbrzymia nawrotowa endometrioza w bliźnie po nacięciu krocza – opis przypadku

Katarzyna Luterek[1], Ewa Barcz[1], Leszek Bablok[1], Zbigniew Wierzbicki[2]

[1] I st Department of Obstetric and Gynecology Medical University of Warsaw, Poland
[2] Department of General Surgery and Transplantology, Transplantation Institute, Medical University of Warsaw, Poland.

Abstract

The occurrence of perineal endometriotic lesions can be explained by mechanical dissemination and transplantation of endometrial cells. Viable decidual endometrial cells are likely to be transplanted into the episiotomy wounds or perineal tears during normal vaginal delivery and subsequent growth may occur. A case of a 33-year old woman with a medical history of recurrent perineal endometriosis was described. An endometriotic giant mass (8 centimeter in diameter) was wide-excised, together with the episiotomy scar. The recovery was uneventful. Three years after the surgery, the patient is symptom-free and with no signs of recurrence or discomfort.

According to the literature and our own experience, a complete excision of endometriotic tissue is the treatment of choice. We can conclude that a wide excision is mandatory as it is the only way to prevent tumor recurrence.

Key words: **endometriosis** / **perineum** / **episiotomy scar** /

Streszczenie

Zmiany endometrialne w obrębie blizny po nacięciu krocza powstają najprawdopodobniej w wyniku transplantacji komórek błony śluzowej macicy, podczas porodu drogą pochwową. W pracy opisano przypadek pacjentki z nawracającą endometriozą w bliźnie po nacięciu krocza. Duży guz o średnicy 8 cm został w całości usunięty chirurgicznie wraz z blizną po nacięciu krocza. Przebieg gojenia był niepowikłany. 3 lata od operacji pacjentka pozostaje asymptomatyczna, bez żadnych dolegliwości. Leczeniem z wyboru w przypadku endometriozy krocza jest szerokie wycięcie guza z marginesem tkanek zdrowych.

Radykalne wycięcie zmiany daje szansę na brak nawrotów. Jest ono również istotne ze względu na przypadki przemiany nowotworowej w obrębie podobnych zmian.

Słowa kluczowe: **endometrioza** / **krocze** / **blizna po nacięciu krocza** /

Adres do korespondencji:
Katarzyna Luterek
I st Department of Obstetric and Gynecology Medical University of Warsaw
Poland, 02-015 Warszawa, Pl.Starynkiewicza 1/3
tel: +48 22 502 14 60; fax: +48 22 502 21 57
e-mail: katarzyna.luterek@wum.edu.pl

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Introduction

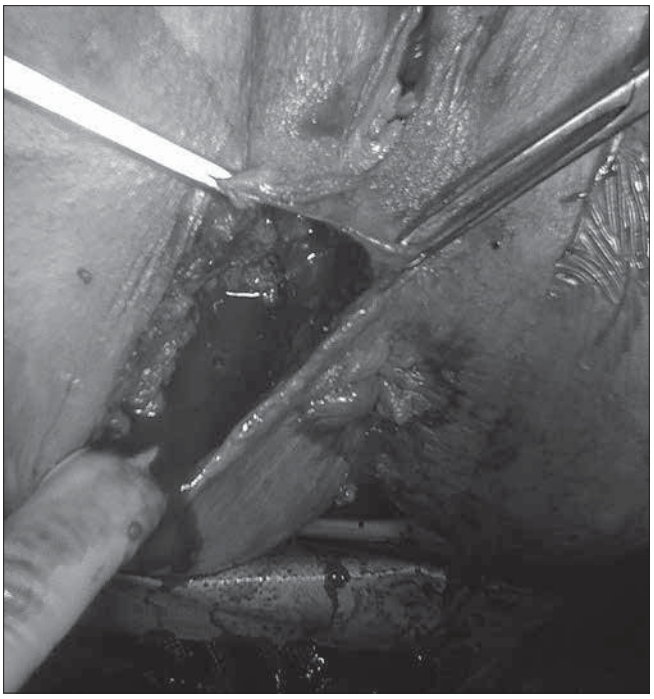
Endometriosis is a condition where functional endometrial tissue is present outside the uterus. The etiopathogenesis of endometriosis is still poorly recognized [1]. Typical signs and symptoms include chronic cyclic dysmenorrhoea, abdominal and pelvic pain, dyschezia (painful defecation), dyspareunia, irregular menstrual bleeding [2]. The estimated prevalence of endometriosis is 5%-15% for women in reproductive period [3]. The disease may affect pelvic as well as extra pelvic locations. It is most commonly observed in the pelvic organs, especially the ovaries, uterosacral ligaments, fallopian tubes, pelvic peritoneum and the Douglas pouch. The extrapelvic localization of endometrial tissue has been described in many organs. Extragenital locations include the intestinal tract and the urinary tract; rarely in inguinal canal, umbilicus, vagina, lungs and the central nervous system [4, 5]. Endometriosis is found in the urinary tract in 1-2% of women suffering from this disease (urinary bladder, ureter) [6]. Endometriosis may occur in surgical scars following laparotomy or laparoscopy, as a result of gynecological and obstetric procedures or general surgery [7, 8, 9].

In the following report we present a case of huge endometriosis (8 cm in diameter) in an episiotomy scar and our approach to evaluation and therapy.

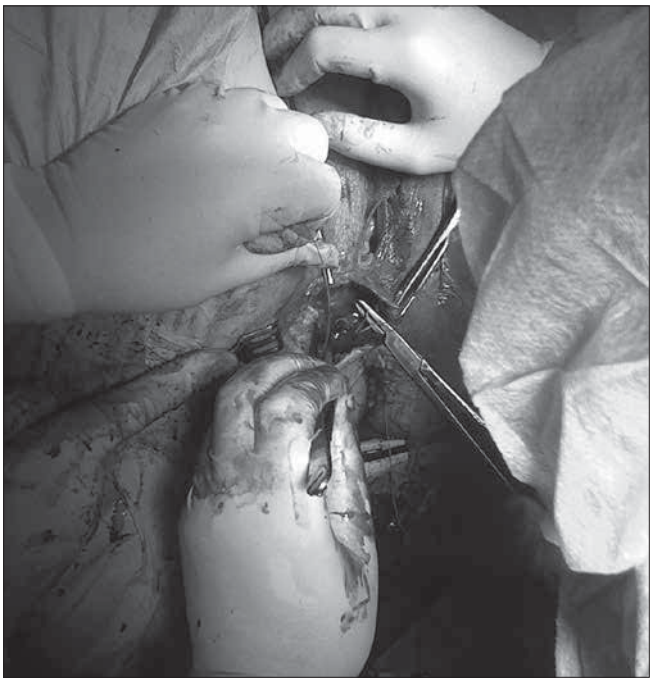
Case history

A 33-year old woman with a medical history of recurrent perineal endometriosis was admitted to the First Department of Obstetrics and Gynecology, Medical University of Warsaw. Her obstetric history was significant for a complicated vaginal delivery in 2000, with prolonged second stage and episiotomy. She delivered a 4400g neonate with birth asphyxia. First symptoms of endometriosis appeared in 2007. The patient presented with perineal pain and a slightly palpable tumor. Revision of the episiotomy scar was performed in a district hospital and an endometriotic-like mass (2 cm in diameter) was excised. Histopathologic examination revealed endometriosis. There was no information about complete excision. The postoperative period was uncomplicated and the patient experienced no pain. Three years later a clinical relapse occurred. Hormonal contraceptives were prescribed at first but no improvement was observed. The pain was correlated with the menstrual cycle and appeared several days before its onset. The patient described it as stabbing, stitching and drawing with dyspareunia. The pain was correlated with tumor growth and was refractory to all forms of analgesics. During physical examination upon admission, a firm nodule of approximately 10.0x 6.0 cm, infiltrating the right buttock, in the contact with the episiotomy scar, was palpated. The infiltration was closely associated with the vagina and reached up to the anal sphincter. The results of gynecological examination (uterus and appendages) and transvaginal ultrasound were normal. Ultrasound trans integument (trans buttock) and trans vaginal imaging demonstrated a multicystic, multicavity change with septa, about 5 cm in diameter. Due to advanced obesity of the patient (BMI 36.9), the ultrasound examination revealed no relation of the node to the urogenital diaphragm. Transanal examination revealed no direct extension to the anal sphincter, rectal and anal wall.

The patient was operated under epidural and general anesthesia in lithotomy position. A team of a gynecologist and a surgeon made a boat-shaped incision in the episiotomy scar, followed by electrosection and scalpel section for complete elimination of the tissue. (Picture 1).



Picture 1. Wide excision with a margin of healthy tissue.



Picture 2. Multilayer sutures on the lodge.

Macroscopic evaluation showed the tumor to be about 8 x 7cm with a typical endometriosis content extracted during preparation. (Picture 3a/b). Following tissue removal, the lodge was sewn up with continuous and type Z stitches (Vicryl Rapid®). (Picture 2). Post-operative wound drainage was performed. Single stitches were placed on the skin wound.

The histopathologic examination revealed numerous, irregular tissue fragments (8x7x4.5 cm in all). Diagnosis – endometriosis external, extragenital. The postoperative course was uneventful, with slight grade anemia (hemoglobin level 87.0 g/L, RBC 3.47 T/L, hematocrit 0.292 L/L). The patient was discharged on the fourth postoperative day. Two years after the surgery, the patient is symptom-free with no signs of recurrence and discomfort.

Discussion

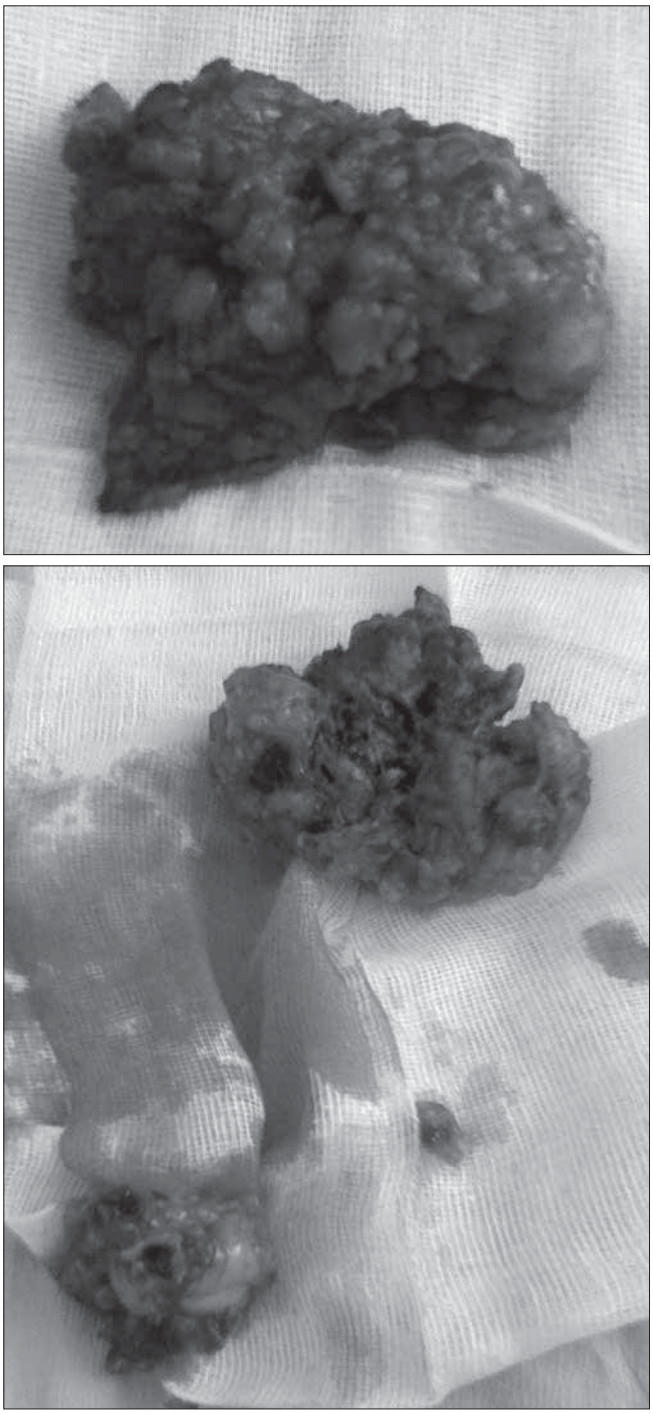
Endometriosis is one of the most common gynecological disorders. Etiopathogenesis of endometriosis has generally been related to endometrial implantation, lymphatic dissemination, coelomic metaplasia and hematogenous spread. Perineal endometriotic lesions can be explained by mechanical dissemination and transplantation of the endometrial cells. During normal vaginal delivery, viable decidual endometrial cells are likely to be transplanted into the episiotomy wounds or perineal tears and subsequent growth may occur. For postoperative wounds, the phenomenon of metaplasia or blood-borne dissemination is less probable.

In the presented case, endometriosis in the perineotomy site seemed to have resulted from implantation during delivery, complicated by damage to the vaginal walls and mucosa. The previous excision had probably been not radical and therefore followed by a relapse in less than a year. Regardless of location, typical symptoms of endometriosis include cyclic occurrence of specific pains. Our patient complained of pain in the right buttock, first experienced a year after the previous endometriosis resection. The pain became more intensive with tumor growth.

Endometriosis can be recognized in physical examination, ultrasonography, MRI. The final diagnosis is made after histopathological examination. MRI is the most sensitive and specific method for diagnosing extraperitoneal endometriosis. It is particularly useful for identification of small changes and differentiation from other integument tumor-like lesions such as a lipoma or an abscess. In the described case no MRI was performed because past medical history indicated recurrence of endometrial tumor in the perineum location. In that case a typical medical interview and ultrasonography were sufficient to diagnose the disease. Physical examination revealed no infiltration of the anal sphincter and the patient was operated in the presence of a proctologist.

The frequency of endometriosis in an episiotomy scar following perineotomy has not been determined. A MEDLINE (1949–2011) search revealed approximately 100 patients who had been reported to have endometriosis in an episiotomy scar. Some of them were found to have involvement of the anal sphincters [10] and a couple had malignant degeneration [8]. Perineal endometriosis published in the literature occurred predominantly as case reports study [11]. Episiotomy is frequently performed at the time of vaginal delivery, whereas endometrial implantation in an episiotomy scar is a relatively uncommon condition. Gunes et al., describe 11 cases of endometriosis in an episiotomy scar following perineotomy with symptoms that appeared, on average, 5-7 years after the delivery and continued for about 9 months from the onset to surgery [12].

In our case, the first symptoms were recorded 7 years after delivery and the first surgery. After tumor excision, the symptoms reoccurred already 12 months later and continued for a period of 2 years.



Picture 3a/3b. Endometriotic mass.

The size of scars following perineotomy is on average 2.5 cm and of tumors following obstetric procedures (cesarean section and perineotomy) is 2.3-3.2 cm. Descriptions of larger tumors are extremely rare and these are mostly lesions in integuments following caesarean sections. To the best of our knowledge, the giant tumor which we described (8x 7.5x 4 cm) is one of the largest ever reported in gynecological literature.

Tumor excision with a margin of healthy tissue is the treatment of choice [13]. The procedure was also performed in our

case due to the fact that it helps to reduce the number of relapses following radical treatment. Surgery is also recommended as a safeguard against malignant transformation.

There are several cases of endometriosis infiltration on the anal sphincter described in literature [10]. In our case no such infiltration was observed either in the rectum or the sphincter, therefore sphincteroplasty was not required. No surgical complications were observed following broad excision of the large tumor.

In the case of our patient we applied hormonal therapy (oral contraceptives) before surgery. Hormonal therapy was discontinued after surgery due to patient obesity (BMI 36,9) and a higher risk of thromboembolic complications. Liang et al., also observed the recurrence of tumor in scar following episiotomy in a patient after surgery with no subsequent hormonal therapy [14]. Hormonal therapy is applied when the tumor is not radically removed or in cases of multiple localizations [11]. Tumor excision is the treatment of choice.

In the period of three years after the surgery no disease recurrence was observed, allowing us to conclude that wide excision is mandatory as the only way to prevent tumor recurrence. Radical wide excision of such large tumor had no unfavorable effect on the functioning of the vagina, anal sphincter and the plastic effect is highly satisfactory.

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