

ABDOMINAL WALL ENDOMETRIOSIS ELEVEN YEARS AFTER CESAREAN SECTION: CASE REPORT

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SUMMARY – Endometriosis is a common chronic disease characterized by growth of the endometrial gland and stroma outside the uterus. Symptoms affect physical, mental and social well-being. Extrapelvic location of endometriosis is very rare. Abdominal wall endometriosis occurs in 0.03%–2% of women with a previous cesarean section or other abdominopelvic operation. The leading symptoms are abdominal nodular mass, pain and cyclic symptomatology. The number of cesarean sections is increasing and so is the incidence of abdominal wall endometriosis as a potential complication of the procedure. There are cases of malignant transformation of abdominal wall endometriosis. Therefore, it is important to recognize this condition and treat it surgically. We report a case of a 37-year-old woman with abdominal wall endometriosis 11 years after cesarean section. She had low abdominal pain related to menstrual cycle, which intensified at the end of menstrual bleeding. A nodule painful to palpation was found in the medial part of previous Pfannenstiel incision. Ultrasound guided biopsy was performed and the diagnosis of endometriosis confirmed. Surgery is the treatment of choice for abdominal wall endometriosis. Excision with histologically proven free surgical margins of 1 cm is mandatory to prevent recurrence. A wide spectrum of mimicking conditions is the main reason for late diagnosis and treatment of abdominal wall endometriosis. In our case, the symptoms lasted for eight years and had intensified in the last six months prior to surgery.

Key words: *Endometriosis – pathology; Abdominal wall – pathology; Cesarean section – adverse effects; Cicatrix; Case reports*

Introduction

Endometriosis is a common chronic disease characterized by growth of the endometrial gland and stroma outside the uterus. It depends on the cyclic change in steroid hormones. Symptoms affect physical, mental and social well-being¹. It occurs in 10%–15% of women of reproductive age, i.e. about 16 million women in the EU. Extrapelvic location of endometriosis is very rare. Abdominal wall endometriosis (AWE) refers to endometrial tissue in the abdominal

wall. It can be primary (umbilical endometriosis) or secondary (after trauma or surgery, when it is called scar endometrioma). Scar endometrioma occurs in 0.03%–2% of women with a previous cesarean section or other abdominopelvic operation. Scar endometrioma is the result of direct implantation of endometrial tissue into the scar at the time of surgery. Differential diagnosis includes abscess, lipoma, hematoma, sebaceous cyst, suture granuloma, incision hernia, lymphoma, etc.². There is an increasing number of cesarean sections and so is the incidence of scar endometrioma as a potential complication of this procedure. There are reports of malignant transformation of AWE^{3,4}. Therefore, it is important to recognize the condition and treat it surgically.

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Case Report

We report a case of a 37-year-old woman presenting with low abdominal pain related to menstrual cycle, which intensified at the end of menstrual bleeding. Her average menstrual cycle duration was 28 days, with five days of vaginal bleeding. She gave birth by cesarean section 11 years before. In her past medical history before cesarean section, no signs of endometriosis were found. The symptoms persisted for the past eight years and aggravated in the last six months. Pain was located in the scar area, irradiating to the pelvis. Considering the present status and patient history of previous cesarean section, AWE was suspected. A nodule painful to palpation was found in the medial part of previous Pfannenstiel incision. Findings of routine laboratory analyses were completely normal, excluding anemia. Transvaginal ultrasound was performed, showing no pathology in the pelvis. Ultrasonographic examination of the abdominal wall showed tumefaction measuring 55x25 mm. Ultrasound guided biopsy was performed. Histopathology of the biopsy specimen revealed fatty and connective tissue infiltrated by focal clusters of siderophages and mononuclear cells together with endometrial glands surrounded by endometrial stroma, which confirmed the diagnosis of endometriosis. Additional work-up was performed. Computerized tomography revealed tumefaction of the abdominal wall above the symphysis, measuring 33x25x38 mm, which was inseparable from the muscle layer. Surgical procedure was recommended. Wide excision in general anesthesia was performed. Histopathologic examination confirmed ectopic endometrial tissue.

Discussion

Endometriosis was first described by Rokitansky in 1860 and it indicates the presence of ectopic growth of endometrial gland and stroma with specific histologic features. There are several theories on the etiology of the disease. The most probable theory is direct implantation of endometrial tissue by transtubal regurgitation during menstrual cycle. Another theory is metaplasia of mesothelial cells in peritoneal cavity under certain conditions. Distant metastases are interpreted by hematogenous and lymphatic spread. There is also a theory of multifactorial polygenic inheritance of endome-

triosis⁵. Direct implantation of endometrial tissue in abdominal wall during surgical procedures is the most probable theory for AWE. Risk factors for endometriosis are early menarche, short menstrual cycles, menorrhagia, infertility, and excessive use of alcohol, caffeine and environmental pollutants¹. Typical locations of endometriotic lesions are the ovaries, pelvic peritoneum, the utero-sacral ligaments and fallopian tubes. Less frequently, endometriosis appears in the lymph, and even more rarely in the colon, other parts of the digestive tract, and the vagina. Very rare locations are urinary tract, lungs, muscles, bones and nervous system. AWE is a rare condition that usually develops in a surgical scar, although it can be found in patients without a history of abdominal surgery (umbilical endometriosis). Diagnosis and removal of AWE is very important. It is a chronic disease and the symptoms can significantly affect the quality of life. Another important reason for surgical treatment is potential relapse or malignant transformation of AWE^{3,4}. The incidence of AWE is 0.03%-2% after cesarean section. However, every surgical procedure in the pelvis carries a certain risk of AWE.

There is a case of AWE in a 15-year-old patient two years after appendectomy and one year after tubo-ovarian abscess⁶. A case of AWE at the site of trocar during laparoscopic removal of ovarian endometrioma has also been reported⁷. Kitajima *et al.* describe a case of AWE in a 26-year-old female with a history of surgery related to bladder extrophy in her neonatal and infantile periods⁸. Kodandapani *et al.* describe a case of umbilical scar endometriosis following laparoscopic-assisted vaginal hysterectomy⁹. One of the most interesting examples of the disease unpredictability is reported by Song *et al.* describing a 9x7.6x6.2 cm large AWE two years after laparoscopic supracervical hysterectomy¹⁰. Spontaneous AWE is rare and makes 9%-20% of all AWEs^{11,12}. Some cases of scar endometriosis have been reported after episiotomy⁷.

The number of cesarean sections is increasing. This could be a reason for the increased incidence of AWE. The triad of mass, pain and cyclic symptomatology helps in the diagnosis, but unfortunately it is not present in all cases¹³. All of these symptoms were present in our case. A wide spectrum of mimicking conditions is the main reason for late diagnosis and treatment of AWE. In our case, the symptoms lasted for eight years and had been intensified in the last six months prior to

surgery. Up to 50% of all cases present without pain related to menstrual cycle^{14,15}. In their study including 65 patients over 12 years, Ecker *et al.* conclude that the primary clinical presentation was abdominal pain in 73.8% and abdominal mass in 63.1% of patients¹². Ozel *et al.* report on abdominal mass in 100% of patients. Pain was present in 83.3% of cases, cyclic in 73.3% and noncyclic in 26.6%¹⁶. A common occurrence is scar augmentation during menstruation. Preoperative diagnosis was accurate in 10%-50% of cases^{15,17}. The most common preoperative diagnosis was neoplasm¹⁸. Differential diagnosis includes abscess, lipoma, hematoma, sebaceous cyst, suture granuloma, hernia, lymphoma and carcinoma.

The ultrasonographic appearance of AWE can widely differ. The most common findings are cystic, polycystic, solid or mixed nodule near to the cesarean section scar with irregular borders, heterogeneous texture characterized by scattered internal hyperechogenic foci, peripheral hyperechogenic ring and scanty vascularity. On computerized tomography, AWE usually appears as a circumscribed solid or mixed mass, enhanced by contrast, often with hemorrhages¹⁹. In our case, after ultrasonography and computerized tomography we performed biopsy that revealed the diagnosis. Surgery is the treatment of choice for AWE. Excision with histologically proven free surgical margins of 1 cm is mandatory to prevent recurrence. To exclude the intraperitoneal spread of the disease in symptomatic patients, some authors recommend explorative laparoscopy^{2,20-22}.

Abdominal wall endometriosis is a rare condition related to prior pelvic surgery. There also are occasional reports of spontaneous AWE. The most common procedure prior to AWE is cesarean section. The number of cesarean sections is increasing, so we can expect an increase in the incidence of AWE. The diagnosis is usually late due to rarity of the condition and specificity of symptoms. Duration of symptoms can significantly influence the quality of life. All these facts call for appropriate and timely diagnosis and treatment of the condition.

References

- Šimunić V. Endometrioza. In: Šimunić V, *et al.*, editors. Reprodukcijska endokrinologija i neplodnost, medicinski potpomognuta oplodnja IVF. Zagreb: Školska knjiga; 2012. p.205-25. (in Croatian)

- Ozturk A, Kaya C, Bozkurtoglu H, Tan N, Yananli ZD, Ucmakli E. Scar endometrioma: an uncommon yet easily treated condition. *J Reprod Med.* 2016;61(5-6):249-53.
- Bats AS, Zafrani Y, Pautier P, Duvillard P, Morice P. Malignant transformation of abdominal wall endometriosis to clear cell carcinoma: case report and review of the literature. *Fertil Steril.* 2008;90(4):1197.e13-6. <http://dx.doi.org/10.1016/j.fertnstert.2007.08.080>
- Stevens EE, Pradhan TS, Chak Y, Lee YC. Malignant transformation of endometriosis in a cesarean section abdominal wall scar: a case report. *J Reprod Med.* 2013;58(5-6):264-6.
- Ciglar S. Endometrioza. In: Kurjak A, *et al.*, editors. Ginekologija i perinatologija. Varaždinske Toplice: Tonimir; 2003. p.612-9. (in Croatian)
- Attia L, Ben Temime R, Sidhom J, Sahli A, Makhoul T, Chachia A, Koubaa A, Zermani R. A case of cutaneous endometriosis developed on an abdominal scar. *Tunis Med.* 2010; 88(11):841-3.
- Emre E, Akbulut S, Yilmaz M, Bozdog Z. Laparoscopic trocar port site endometriosis: a case report and brief literature review. *Int Surg.* 2012;67(2):135-9. <http://dx.doi.org/10.9738/CC124.1>
- Kitajima T, Inoue M, Uchida K, Otake K, Kusunoki M. Scar endometriosis in a patient with bladder extrophy. *Int Surg.* 2013;98(2):145-8. <http://dx.doi.org/10.9738/INTSURGD-12-00011.1>
- Kodandapani S, Pai MV, Mathew M. Umbilical laparoscopic scar endometriosis. *J Hum Reprod Sci.* 2011;4(3):150-2. <http://dx.doi.org/10.4103/0974-1208.92291>
- Song JY, Borncamp E, P Mehaffey, Rotman C. Large abdominal wall endometrioma following laparoscopic hysterectomy. *JLS.* 2011;15(2):261-3. <http://dx.doi.org/10.4293/108680811X13071180407078>
- Papavramidis TS, Sapalidis K, Michalopoulos N, Karayanopoulou G, Raptou G, Tzioufa V, Kesisoglou I, Papavramidis ST. Spontaneous abdominal wall endometriosis: a case report. *Acta Chir Belg.* 2009;109(6):778-81.
- Ecker AM, Donnellan NM, Shephard JP, Lee TT. Abdominal wall endometriosis: 12 years of experience at a large academic institution. *Am J Obstet Gynecol.* 2014;211(4):363.e1-5. <http://dx.doi.org/10.1016/j.ajog.2014.04.011>
- Dell'Acqua S, Colosi E, Angiolollo M, Rivela G, Bovenga S, Natale A. Endometriosis of the abdominal wall after cesarean section. *Minerva Ginecol.* 1993;45(6):327-31.
- Đorđević M, Jovanović B, Mitrović S, Đorđević G, Radovanović D, Sazdanović P. Rectus abdominis muscle endometriosis after cesarean section – case report. *Acta Clin Croat.* 2009;48(4): 439-43.
- Bektas H, Bilsel Y, Sari YS, Ersoz F, Koc O, Deniz M, Boran B, Hug GE. Abdominal wall endometrioma: a 10-year experience and brief review of the literature. *J Surg Res.* 2010;164:e77-81. <http://dx.doi.org/10.1016/j.jss.2010.07.043>
- Ozel L, Sagioglu J, Unal A, Unal E, Gunes P, Baskent E, Aka N, Titiz MI, Tufekci EC. Abdominal wall endometriosis in the cesarean section surgical scar: a potential diagnostic pitfall.

- J Obstet Gynaecol Res. 2012;38(3):526-30. <http://dx.doi.org/10.1111/j.1447-0756.2011.01739.x>
17. Patterson GK, Winburn GB. Abdominal wall endometriomas: report of eight cases. *Am Surg*. 1999;65(1):36-9.
18. Heller DS, Fitzhugh VA. Abdominal wall endometriosis: a rarely anticipated diagnosis: a 16-year experience and brief literature review. *J Reprod Med*. 2014;59(3-4):110-2.
19. Eljuga D, Klarić P, Bolanča I, Grbavac I, Kuna K. Abdominal wall endometriosis: case report. *Acta Clin Croat* 2012;51(2):261-2.
20. Mistrangelo M, Gilbo N, Cassoni P, Micallef S, Faletti R, Miglietta C, Brustia R, Bonnet G, Gregori G, Morino M. Surgical scar endometriosis. *Surg Today*. 2014;44(4):767-72. <http://dx.doi.org/10.1007/s00595-012-0459-3>
21. Miccini M, Gregori M, Ferraro D, Ciardi A, Cassibba S, Biacchi D. Abdominal scar endometriosis: case report. *Clin Exp Obstet Gynecol*. 2016;43(3):431-3.
22. Gupta P, Gupta S. Scar endometriosis: a case report with literature review. *Acta Med Iran*. 2015;53(12):793-5.

Sažetak

ENDOMETRIOZA TRBUŠNE STIJENKE JEDANAEST GODINA NAKON CARSKOG REZA: PRIKAZ SLUČAJA

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Endometrioza je česta kronična bolest obilježena rastom endometrijskih žlijezda i stome izvan maternice. Simptomi utječu na pad fizičkog, mentalnog i socijalnog blagostanja. Ekstrapelvična lokacija endometriozne je rijetka. Pojavnost endometriozne trbušne stijenke je 0,03%-2% žena s prethodnim carskim rezom ili nekom drugom abdominopelvičkom operacijom. Vodeći simptomi su abdominalna nodularna masa, bol i ciklička simptomatologija. Broj carskih rezova je u porastu pa tako raste i pojava endometriozne trbušne stijenke kao potencijalne komplikacije ovoga zahvata. Opisani su slučajevi maligne transformacije endometriozne trbušne stijenke. Zato je potrebno pravodobno prepoznati ovo stanje i liječiti ga kirurški. Prikazan je slučaj 37-godišnje bolesnice s endometriozom trbušne stijenke 11 godina nakon carskog reza. Imala je bolove u donjem dijelu trbuha koji su bili povezani s menstruacijskim ciklusom i pojačavali su se prema kraju menstrualnog krvarenja. Bolni čvorčić se pipao u srednjem dijelu ožiljka od carskog reza. Ultrazvučno navođena biopsija je potvrdila dijagnozu endometriozne. Metoda izbora u liječenju endometriozne trbušne stijenke je kirurški zahvat. Široka ekscizija s patohistološki dokazanim slobodnim rubom od 1 cm je obvezna kako bi se spriječio recidiv endometriozne. Široka diferencijalna dijagnoza je razlog kasne dijagnoze i liječenja endometriozne trbušne stijenke. Simptomi su trajali osam godina, a pojačali su se šest mjeseci prije operacije.

Ključne riječi: *Endometrioza – patologija; Trbušna stijenka – patologija; Carski rez – štetna djelovanja; Ožiljak; Prikazi slučaja*