

## RECTUS ABDOMINIS MUSCLE ENDOMETRIOSIS AFTER CESAREAN SECTION – CASE REPORT

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**SUMMARY** – Endometriosis is defined by the presence of functional endometrial tissue outside the uterus, where it is normally located. Endometriosis is one of the most common gynecologic entities affecting 8%-18% of menstrual women. Endometriosis can occur at intra- and extrapelvic localizations. The most common intrapelvic localizations are those involving the ovaries, Douglas' area, pelvic peritoneum, uterus, bladder and rectum. Abdominal endometriosis is the most common localization of extrapelvic endometriosis and usually develops in connective tissue. Extra-pelvic implantation of endometrial tissue may develop in any organ including the skin, lungs, liver, extremities, brain and stomach. Three years after cesarean section, a 35-year-old female was operated on for suspected anterior abdominal hernia at the site of previous section. An egg-sized tumor was removed from the rectus abdominis muscle and referred for histopathologic and immunohistochemical analyses. The results showed endometriosis of the muscle with positive estrogen and progesterone receptors. A year after the procedure, treatment with gonadotropin-releasing hormone analogs was continued due to recurrent pain in the scar area, along with ultrasonography and biochemical marker (carbohydrate antigen 125) follow-up. Clinical diagnosis of scar endometriosis can be made by thorough history and physical, ultrasonography and biochemical examinations. Scar endometriosis should always be considered when the symptoms occur in a cyclic and hormone-dependent pattern, mostly after gynecologic operations, and worsening during menstruation. Definitive diagnosis is based on histopathologic analysis.

**Key words:** *Cesarean section – adverse effects; Endometriosis – diagnosis; Endometriosis – etiology; Endometriosis – therapy; Cicatrix – etiology; Case report*

### Introduction

Endometriosis is defined by the presence of functional endometrial glands and stroma outside the uterus, where they are normally located. Endometriosis mainly occurs during fertile age. It is diagnosed in women aged 25-30, in infertile women and/or those

with pelvic pains in particular. It is by no means infrequent in adolescents. Endometriosis is diagnosed in nearly half of women aged <20 that suffer chronic pelvic pains and/or dyspareunia. About 5% of endometriosis cases are found in postmenopausal women, where risk factors are hormone replacement therapy, body mass index (BMI) >30 and causal hyperestrogenism as a result of the increased androstenedione conversion to estrone. Rarely, the disease is diagnosed in men on long term estrogen therapy<sup>1-5</sup>.

Endometriosis is one of the most common gynecologic entities, present in 8%-18% of menstrual women

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and is characterized by implantation of ectopic or heterotopic endometrial tissue that reacts to hormone stimulation. Endometriosis is incidentally found in over 20% of gynecologic laparoscopies and is clinically presented by menstrual irregularity, dysmenorrhea, dyspareunia, infertility and frequent symptoms of acute abdomen. Endometriosis is the third reason for hospitalization for gynecologic problems in the USA<sup>4-6</sup>.

Endometriosis can be of intra- or extrapelvic localization. Intrapelvic endometriosis is the most common localization, mostly presenting in the ovaries, Douglas' area, pelvic peritoneum, uterus connections, urinary bladder, rectum, etc.<sup>7</sup>.

Extrapelvic implantation of endometrial tissue can be found in other organs of the body, e.g., in the lungs, liver, extremities, brain, abdomen and episiotomy, including the skin. Recent studies have provided definitive evidence for the increasing risks of breast cancer, ovary cancer, melanoma and non-Hodgkin's lymphoma in women with endometriosis. These women and their closest relatives also have a higher incidence of thyroiditis, hypo- and hyperthyroidism, autoimmune disease such as rheumatic arthritis, lupus, sclerosis and Meniere's disease, which undoubtedly points to the role of the immune component in the development of endometriosis<sup>8-10</sup>.

### Case Report

A 35-year-old woman from Novi Pazar was admitted to University Department of Obstetrics and Gynecology, Kragujevac Clinical Center, on March 7, 2008, for the treatment of endometriosis of rectus abdominis muscle. The patient had a female phenotype. Pubic distribution of hairiness and secondary sexual characteristics were normal. An infraumbilical scar was visible. Gynecologic history revealed menarche since the age of 16, with irregular menstrual cycle lasting for 15-20 days and total 5-day duration of menstruation. She denied earlier gynecologic diseases. She had two pregnancies, eight and five years before, both terminated by cesarean section because of pregnancy induced hypertension. She had two full-term pregnancies and delivered two children of over 2750 g birth weight.

Three years after the last cesarean section, she felt pain in the abdominal wall, and several months later

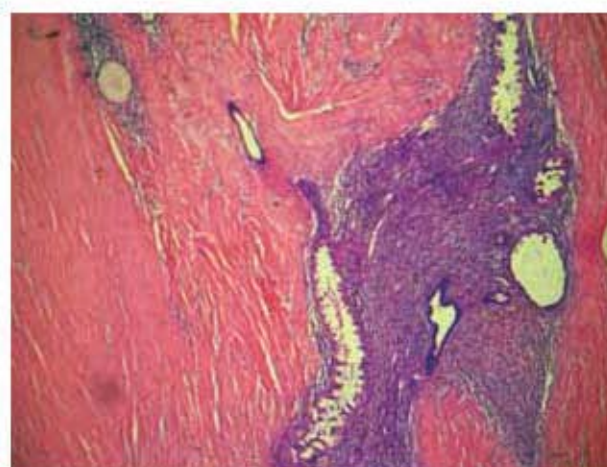


Fig. 1. Endometriosis of rectus abdominis muscle (HE, X40).

this discomfort transformed to swelling. The patient underwent operation by a surgeon on February 7, 2007, for suspicion of abdominal wall hernia at the site of surgical incision. An egg-sized tumor was removed from the muscle and referred for histopathologic analysis (Fig. 1). A gynecologist was not consulted, since histopathologic analysis took 20 days and the patient was discharged from Department of Surgery.

### Histopathologic findings

Macroscopically, the tissue sample had 60x40x30 mm in dimensions, rough surface, covered with bare greasy tissue, of solid consistency, white in color on

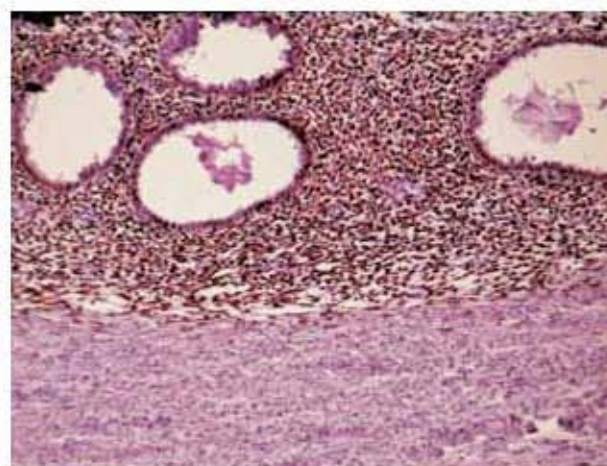


Fig. 2. Endometriosis of rectus abdominis muscle; progesterone receptors (X100).

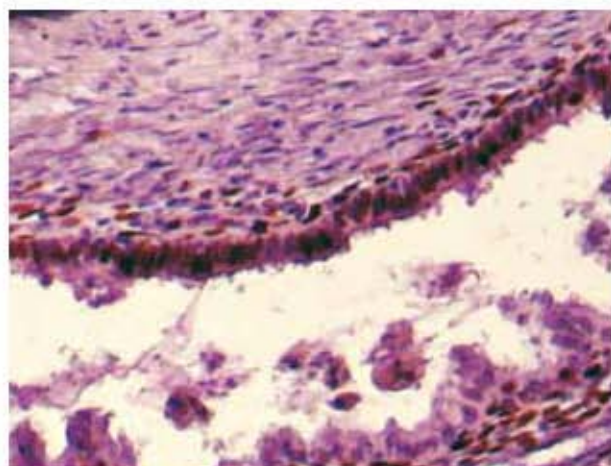


Fig. 3. Endometriosis of rectus abdominis muscle; estrogen receptors (X200).

section. Microscopically, gland-like formations were present on serial sections of the sample analyzed, between the muscle and binding threads. These formations were overlain with one line of columnar epithelium, while the formations were encircled with multiple stromal cells, which corresponded to ectopic localization of endometrial mucous membrane. Immunohistochemically, the epithelium and stromal cells of the endometriosis focus expressed estrogen and progesterone receptors (Figs. 2 and 3). Specific biochemical Masson trichrome staining proved it to be striated muscle (Fig. 4).

After the operation, neither follow-up by the carbohydrate antigen (CA 125) biochemical marker was

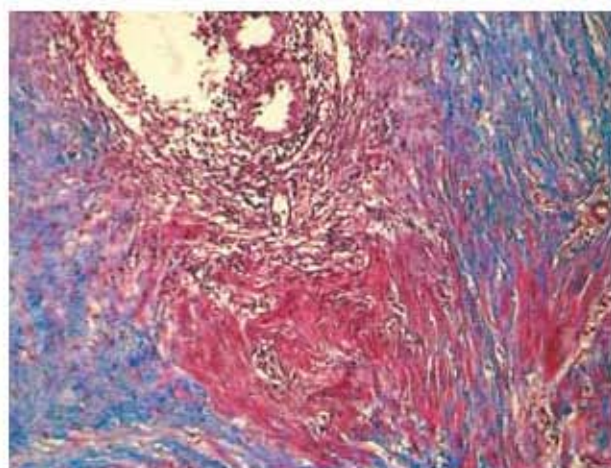


Fig. 4. Endometriosis of rectus abdominis muscle (Masson stain, X100).

performed nor therapy with gonadotropin-releasing hormone (Gn-RH) analogs was continued at primary health care level.

During the last two months and a year after re-laparotomy performed by the surgeon, the patient began to feel mild pains at the same site, now not connected with menstrual cycle. Palpation and ultrasonography showed no changes at the site of incision. Due to suspected recurrence of endometriosis, the CA 125 biochemical marker (56 IU/mL) and therapy with Gn-RH analogs in six cycles was administered. The follow-up of therapeutic effects, CA 125 marker and ultrasonography studies are under way.

## Discussion

Endometriosis is not a disease only reserved for humans, as it can also be found in other primates. Endometriosis was first described by Rokitsansky in 1860 as the presence of endometrial tissue outside the uterus<sup>10,11</sup>.

Endometriosis outside the pelvis is a relatively unknown disease, accounting for 12% of all endometriosis cases. Endometriosis of the abdominal wall is the most common localization of extrapelvic endometriosis, usually developing in an old surgical scar; however, cases of spontaneous endometriosis have also been described. It is most often found after gynecologic and obstetric operations, such as cesarean section, hysterotomy for different indications and hysterectomy; cases after early amniocentesis have also been reported<sup>10-12</sup>.

Almost always there is a dilemma concerning accurate diagnosis. The use of ultrasonography, computerized tomography and nuclear magnetic resonance can be helpful. On differential diagnosis, diseases such as sarcoma, ovary cancer, melanoma, lymphoma, hernia, hematoma and abscesses can also result in surgical operation<sup>1,13</sup>.

In our case, the patient consulted the surgeon because of the change in the abdominal area. Then, it was thought to be ventral hernia that developed three years after the last cesarean section, when laparotomy was performed by low longitudinal incision. Some studies have shown that endometriosis develops on the scar three months after the operation to up to ten years, most likely within 18 months<sup>7,14</sup>.

Changes on the scar can be solid, cystic, or combined. In our case, the change was egg-sized and solid

in structure. Other authors indicate that these changes mostly have a few centimeters in diameter. Because of the possible recurrence, wide excisions are needed. Other preventive measures are also warranted, including precise operative techniques on cesarean section, abdomen lavage, etc. Relapses are extremely rare but they have been recorded.

Microscopic analysis of the removed tissue revealed a number of gland formations, with their own stroma. These formations were scattered across the connective and muscle fibers. The gland lumina were coated with a layer of columnar epithelium and were surrounded by multiple stromal cells. The finding of striated muscle fibers pointed to endometriosis of the rectus abdominis muscle<sup>1,15</sup>.

Endometriosis as an ectopic collection reacts to hormonal influences as a hormone-dependent tissue. These collections contain endometrial stroma and gland elements, which can react decidually and as uterine mucosa. In our case, immunohistochemical analysis showed the nuclei of epithelial and stromal cells to express estrogen and progesterone receptors. Progesterone receptors in stromal cell nuclei showed higher positivity than estrogen receptors in epithelial cells. A strongly positive receptor staining for estrogen and progesterone in the epithelial and stromal cell nuclei indicated the tumor to show hormonal response connected with menstrual cycle; this reaction is identical to that of uterine endometrium. Due to recurrent pain in the scar, the patient was referred for gynecological treatment. Gn-RH analog therapy reduced ovarian function and led to hypophyseal hypogonadism that resulted in atrophy of ectopic epithelium<sup>16,17</sup>.

An explanation for scar endometriosis corresponds mostly to the mechanical transplantation theory. To prove this, endometriosis occurs mainly after gynecologic operations. After cesarean section, the incidence is 0.03% to up to 0.45%. The reason for the highest incidence of endometriosis after cesarean section is that endometrial tissue during pregnancy is most suitable for transplantation. This theory needs a tenable transfer of endometrial tissue to another location where it can grow and proliferate, and this can be done with surgical instruments. As an alternative there is a hypothesis that mesenchymal cells may differentiate to endometrium. Some scientists provide a combined explanation that endometrium transport to the ectopic

site occurs first, and then ectopic cells induce metaplasia of the surrounding cells into the endometrial tissue. On the other hand, pregnancy reacts protectively to the occurrence of endometriosis<sup>5,16,18</sup>.

In general, endometriosis is found on the skin, on the very scar, or adjacently to it. The next most often localization is subcutaneous endometriosis. Endometriosis of rectus abdominis muscle is extremely rare and it has been described as a single case. Scar endometriosis is rarely found after appendectomy, cholecystectomy and other abdominal operations or laparoscopy. However, post-laparoscopy endometriosis has lately being on an increase because the method is often used for pelvic endometriosis, which is probably the reason for its occurrence after other operations. It is interesting that scar endometriosis is rarely found together with pelvic endometriosis. Episiotomy endometriosis is also rarely reported<sup>19,20</sup>.

## Conclusion

In general, scar endometriosis can be diagnosed clinically by careful personal medical history, disease history and physical examination. Scar endometriosis should always be considered when symptoms occur in a cyclic manner, mainly after gynecologic operations, and when these symptoms worsen during menstruation. Ultrasonography and CA 125 biochemical marker follow-up can be useful. The accurate diagnosis is reached by histopathology. Due to the increasing rate of cesarean sections, gynecologic operations, laparoscopies and amniocentesis, an increasing incidence of extrapelvic localization of endometriosis is expected.

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### Sažetak

#### ENDOMETRIOZA RAVNOG TRBUŠNOG MIŠIĆA POSLIJE CARSKOG REZA – PRIKAZ SLUČAJA

M. Đorđević, B. Jovanović, S. Mitrović, G. Đorđević, D. Radovanović i P. Sazdanović

Endometrioza se definira prisustvom funkcionalnih endometrijalnih žlijezda i strome izvan šupljine maternice, gdje je njihova normalna lokalizacija. Endometrioza je jedan od najčešćih ginekoloških entiteta, prisutna kod 8%-18% menstrualnih žena. Endometrioza može biti intra- i ekstrapelvične lokalizacije. Najčešći slučajevi su intrapelvične lokalizacije koje zahvaćaju ovarijume, zatim Douglasov prostor, pelvični peritoneum, veze maternice, mokraćni mjehur, rektum. Abdominalna endometrioza je najčešća lokalizacija ekstrapelvične endometrioze i obično se razvija na vezivnom tkivu. Ekstrapelvična implantacija endometrijalnog tkiva može biti na bilo kom organu u organizmu uključujući i kožu, epiziotomiju, pluća, jetru, ekstremitete, mozak i trbušni zid. Pacijentica stara 35 godina tri godine poslije carskog reza operirana je zbog pretpostavke da se radi o kili prednjeg trbušnog zida, na mjestu prethodnog reza, kada je odstranjen tumor veličine jajeta na ravnom trbušnom mišiću. Histopatološka i imunohistokemijska analiza tumora pokazala je da se radi o endometriozi dotičnog mišića sa pozitivnim estrogenskim i progesteronskim receptorima. Godinu dana poslije procedure pojavila se bol u predjelu ožiljka, bolesnica je stalno liječena analogom hormona za otpuštanje gonadotropina, uz serijsku kontrolu ultrazvuka i biokemijskog markera, CA 125. Klinička dijagnoza ožiljne endometrioze može se postaviti pažljivom anamnezom i fizikalnim pregledom, ultrasonografskim i biokemijskim ispitivanjem. Na ožiljnu endometriozu treba misliti uvijek kada se simptomi javljaju ciklično, u zavisnosti od hormona, najčešće poslije ginekoloških operacija, a koji se pogoršavaju tijekom menstruacije. Prava dijagnoza postavlja se histopatološkom analizom.

Ključne riječi: *Carski rez – štetni učinci; Endometrioza – dijagnostika; Endometrioza – etiologija; Endometrioza – terapija; Ožiljak – etiologija; Prikaz slučaja*