

# Scar endometriosis, a tricky diagnosis.

## A case series report and a short review of the literature



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Nikoletta Rapti<sup>\*/°°</sup>, Dionysios Katsaounis<sup>\*\*</sup>, Georgios Rallis<sup>°</sup>, Stella Ioannou<sup>\*\*/°°</sup>,  
Dimosthenis Rammos<sup>°°</sup>, Dimitrios Filippou<sup>°°</sup>, Panagiotis Skandalakis<sup>°°</sup>

<sup>\*</sup>*Surgical Department, Argos General Hospital, Argolida, Greece*

<sup>\*\*</sup>*Surgical Department, General Hospital of Nikaia, Piraeus, Greece*

<sup>°</sup>*Surgical Department, General Hospital of Athens "Elpis", Athens, Greece*

<sup>°°</sup>*Department of Anatomy and Surgical Anatomy, Medical School, National and Kapodestrian University of Athens, Athens, Greece*

### Scar endometriosis, a tricky diagnosis. A case series report and a short review of the literature

**BACKGROUND:** *Abdominal wall endometrioma is a rare condition, which usually develops in surgical scar of a Cesarean section (C-section) or of a hysterectomy. Scar endometriosis is a misdiagnosed condition in general surgery, since it mimics other surgical conditions. Diagnosis is mainly based upon a high index of suspicion.*

**AIM:** *The purpose of this study is to highlight the characteristics of this rare pathology and assess the diagnostic algorithm and therapeutic options.*

**METHODS:** *The article is a case series and therefore no specific methods have been applicable.*

**RESULTS:** *In the current study, we present a case series of seven cases with abdominal pain on the Pfannenstiel incision, treated in our department during a two years period, between 2014 and 2016, followed by a brief review of the literature.*

**CONCLUSION:** *Scar endometriosis may be difficult to diagnose as it is an unfamiliar entity to general surgeons. This condition can be confused with other surgical conditions. Imaging methods, such as ultrasound or CT-scan should be used for differential diagnosis. The prevention of scar endometriosis may also be important. Surgical excision is the treatment of choice for scar endometriosis.*

**KEY WORDS:** Abdominal wall, Caesarian, Endometriosis, Scar

### Introduction

Endometriosis is the condition in which there is a functional endometrium outside the uterine cavity that responds to hormonal changes of the menstrual cycle <sup>1,2</sup>. Ectopic endometrial tissue usually is located in the pelvis, but also can be found associated with the lungs, bowel, ureter, extremities and abdominal wall. Regarding the extra-pelvic sites the most frequent one is the abdomi-

nal wall; endometrial tissue has been identified in the skin, subcutaneous tissues and abdominal muscular layers and nowadays it is closely related to numerous procedures. Most commonly it is a result of gynecological or obstetrical operations, however scar endometriosis is a rare site <sup>1,2</sup>.

General surgeons often deal with such occurrences because abdominal wall endometriosis mimics other surgical conditions, such as postoperative hernia <sup>3</sup>. This entity can result in unnecessary procedures, delayed or misdiagnosis and can cause emotional and physical distress to the patient. In the current study we present a case series of scar endometriosis treated in our department, followed by a brief review of the literature, with an aim to elucidate the main clinical features in order to create a high index of suspicion between surgeons.

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*Correspondence to: Nikoletta Rapti, M.D, Surgical Department, Argos General Hospital, Argolida, Korinthou str. 191, GR-21231 Argos, Greece (e-mail: nicoledoc@gmail.com)*

## Case Reports

### CASE N. 1

A 37-year-old patient presented to the emergency department with abdominal pain on the Pfannenstiel incision. The patient was in the middle of her menstrual cycle. Her personal history did not include any chronic disease, she did not take any medication, she did not mention any known allergies and, according to her surgical history, she had undergone a C-section 6 years ago and a second C-section with a plastic repair of postoperative hernia 5 years ago. After the first operation she reported periodic pain at the left side of the Pfannenstiel incision during the middle of her menstrual cycle. The physical examination found a palpable and painful formation of 2x3cm on the left edge of the Pfannenstiel section with no other pathological findings. Laboratory findings were normal. Ultrasound examination that followed showed a hypervascularized subcutaneous formation of 2x3 cm. Both clinical and imaging findings lead the patient to the operating theater, where a fibrous structure was identified and removed entirely, along with healthy surrounding tissue. Intraoperative findings suggested an endometrial tissue which was histologically confirmed. The patient had a normal postoperative course and was discharged the next day completely asymptomatic.

### CASE N. 2

A 36-year-old female, who underwent cesarean section 3 years back, presented with a painful lesion at the Pfannenstiel incision for the last year. Her personal history was free. Her symptoms had a cyclic character, accompanying her menstruation. She experienced cycle length variations. On her abdominal examination a painful lesion of about 2x3cm was found at the middle of the stich line. The ultrasound examination revealed a circumscribed hypervascularized formation. The patient was subjected to surgery. A mass 3x2x2cm

was found above the rectus sheath and was widely excised (Fig. 1). The histopathological report confirmed it to be endometriosis.

### CASE N. 3

A 34-year-old female was admitted to our department, a year following cesarean section due to a painful lesion at the left margin of the surgical scar. Her personal history was unmentioned. She complained for acute pain during her menstruation. The physical examination revealed probably two different masses medial to the left margin of her scar. An abdomen MRI was performed and showed two masses on the rectus abdominis muscle, 4.5x3.5x2cm and 2.7x1.6x1.3cm, with high signal intensity on T1W and low on T2W. The MRI scan diagnosis was endometriosis. The patient underwent a wide excision of both masses and because of the large deficit of the muscle an intraperitoneal mesh was used to close it. The histopathological report confirmed it to be endometriosis. The patient had a normal postoperative course and was discharged the next day completely asymptomatic.

### CASE N. 4

A 33-year-old woman presented to our department with the complaints of pain and swelling on the upper part of cesarean scar for the last 3 years. She had undergone 2 cesarean deliveries in the past, 5 years and 3 years ago. Her personal history was unmentioned. On her abdominal examination a small lesion of about 2x2cm was found under the umbilicus laterally. The ultrasound revealed a hypoechoic subcutaneous mass. A contrast-enhanced CT scan showed a mass 2x3cm on the rectus sheath without pathological enhancing. She underwent a wide excision of the mass and a part of the anterior laminae. For better closure a prolene mesh was used with the sublay technique. The histopathological report confirmed it to be endometriosis (Fig. 2).

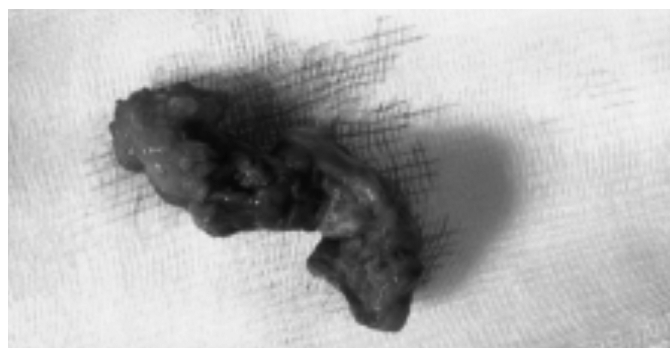


Fig. 1: Resected endometrioma.

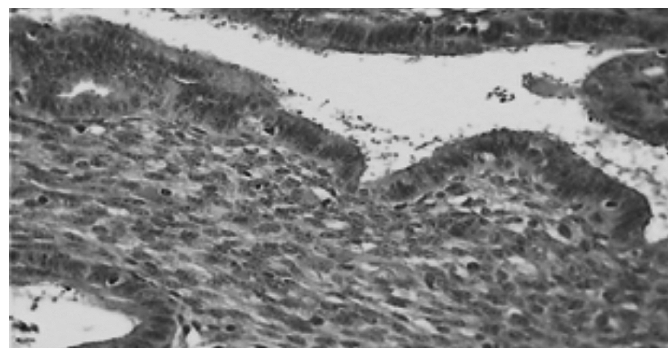


Fig. 2: Endometrial glands and stroma within fibrous tissue.

TABLE I - Synopsis of reported patients

Age, y	C/s	years after C/s	Periodical symptoms	u/s	CT scan	MRI scan	Recurrence
37	2	6	yes	+	-	-	No
36	1	3	yes	+	-	-	No
34	1	1	yes	-	-	+	No
33	2	5	no	-	+	-	No
27	1	4	no	+	-	-	No
29	1	3	no	+	-	-	No
32	2	3	no	+	-	-	No

CASE N. 5

A 27-year-old woman was seen in the outpatient surgery clinic with the complaints of pain at the right margin of cesarean scar for the last 4 years, which was initially present at the time of her menstrual cycle, but later became continuous in nature. According to her surgical history, she had undergone an orthopedic surgery on her left knee due to trauma and a C-section 4 years ago. The physical examination did not reveal something unusual. The ultrasound showed a hypoechoic subcutaneous mass 2x1cm at the right margin of her scar. As a diagnosis of stitch granuloma was made, the patient was taken up for excision of the mass under local anesthesia histopathological examination revealed it to be a case of scar endometriosis.

CASE N. 6

A 29-year-old female was admitted to our department, three years following cesarean section due to a painful lesion on the surgical scar. Her personal history was unmentioned. Examination revealed 2x2 cm mass at the upper part of the cesarean scar. Ultrasound examination showed a hypoechoic mass surrounded by a hyperechoic ring of variable width, and vascularity was present. The patient was underwent a wide excision under local anesthesia. The histopathological report confirmed it to be endometriosis.

CASE N. 7

A 32-year-old female was admitted to our department with a painful mass on the right margin of her Phannestiel incision, 9 months after her last cesarean section. She had undergone 2 cesarean deliveries in the past, 3 years and 9 months ago. The ultrasound findings were a hypoechoic mass with vascularity. The patient was underwent a wide excision under local anesthesia. The

histopathological report confirmed it to be endometriosis Table I.

All the seven patients were followed up for a period of 1 year following the operation with no signs of recurrence. All patients were seen, and diagnosed between January 2012 and December 2014.

Discussion

Endometriosis mainly concerns women in the reproductive age and its prevalence is reported to be 8-15% for these women <sup>1</sup>. Endometrial tissue has been found in many sites. Most commonly it is found in the pelvis, however extrapelvic positions have been described such as lungs, urinary tract, bowel, and abdominal wall. Cecal localization of endometriosis is the rarest site among the large intestine, however it might mimics acute appendicitis in the emergency setting <sup>4</sup>. Endometriosis of the abdominal wall may also arise in the absence of a history of surgical operations (20% of patients) <sup>5</sup>.

Scar endometriosis is a rare position. Specifically, the incidence after a hysterectomy is under 2%, while after a C-section it is 0.03-0.4% <sup>[5-7]</sup>. More rare are postoperative occurrences related to surgery on the fallopian tubes, appendectomy and amniocentesis <sup>6</sup>. Extremely rare is endometriosis of the uterus incision, but it is referred in the bibliography <sup>8</sup>. The time between the surgical operation and the clinical occurrence varies from 3 months to 10 years <sup>9</sup>.

The pathogenesis of endometriosis is not completely known. It is thought that there are predispositional genetic factors that combined with other immunological and biochemical factors result to the generation of endometriotic foci outside the uterus. There are various theories trying to interpret the pathogenesis of endometriosis in the literature. Sampson was the first to describe the theory of regression and implantation of endometrial tissue during menstruation in the pelvic structures <sup>10</sup>. According to this theory we can explain the scar endometriosis. It is thought to be generated

from direct implantation of endometrial cells in the fascias of the abdominal wall or in the subcutaneous tissue during the surgical operation. This theory is supported by experiments where the normal menstruation is spread in the abdominal wall, thus creating an endometriotic focus. Nevertheless, this theory does not explain the rare endometriosis cases in organs such as lungs, kidneys or brain. Halban developed the theory of vascular spreading. According to that, endometrial cells find their way to the vascular circulation through lymphovascular channels and are thus transferred to ectopic sites<sup>11</sup>. A third theory refers to an abdominal wall cells metaplasia into endometrial tissue. This metaplasia is generated either from a displacement or from hormonal stimulation<sup>12</sup>. There are some researchers that support a combination of the above theories.

Symptoms are not always typical and are present in about 20% of patients with scar endometriosis. The most common symptom is the local periodic pain combined with local palpable mass, present in the middle of the menstrual cycle<sup>1</sup>. The key point for the clinical diagnosis is the circular nature of the signs and symptoms of the patient. Differential diagnosis includes postoperative hernia, lipoma, hematoma, fat necrosis, abscess, and tumors such as desmoid tumors, lymphomas, sarcomas, skin malignancies and metastatic tumors<sup>3</sup>. Horton et al state that the accurate preoperative diagnosis varies between 20-50%. A possible explanation for this diagnostic failure is that most times the diagnosis had to be made by the general surgeon who is not familiar with that disease. Another diagnostic trap is the nonspecific clinical presentation of scar endometriosis and the number of possible diagnosis. Malignant transformation of scar endometriosis is rare (0.3-1%) with a pathogenetic mechanism still unknown<sup>13</sup>. Local invasion is a common dissemination way, but few cases of lymph nodes metastases are described in the literature<sup>13</sup>. Histology of extra-ovarian transformations are primarily represented by endometrioid carcinoma (69%), followed by sarcoma (25%), clear cell carcinoma (4.5%), and other variants (4.6%)<sup>14</sup>. The simultaneous occurrence of pelvic endometriosis with scar endometriosis is infrequent<sup>15</sup>. A routine laparoscopic pelvic examination is not recommended<sup>2</sup>.

Diagnosis is mainly based on clinical findings. Additional work up includes ultrasound, color Doppler sonography, CT scan and MRI. Ultrasonography is the best and most commonly used diagnostic modality for abdominal masses. The scar endometriotic focus appears as cystic or polycystic, mixed or solid mass, with its vasculature as depicted in the color Doppler, but all these findings are nonspecific<sup>16,17</sup>. The CT scan is also nonspecific. Endometriosis has no pathognomonic findings on CT, as imaging depends on the phase of the menstrual cycle. MRI can be useful for preoperative estimation of deep pelvic endometriosis, of endometriosis of the abdominal wall muscles and of the subcutaneous tissue<sup>18,19</sup>. MRI

can locate minor lesions and detect the hemorrhagic sign of endometriosis<sup>20</sup>. Fine needle aspiration cytology (FNAC) could be diagnostic and exclude the possibility of malignant transformation, but it should not be used when a postoperative hernia is suspected. Its use is controversial because there is the possibility of a new implantation<sup>21</sup>. Moreover some studies show that FNAC is not diagnostic in patients with scar endometriosis<sup>22</sup>.

The therapeutical options for section endometriosis are conservative and surgical. The conservative treatment includes contraceptive pills, danazol and GnRH. The hormonal treatment inhibits the oestrogen composition and generates the atrophy of ectopic endometrial foci, as well as the suspension of circular stimulation and their hemorrhage. The response is partial and unfortunately there is a high possibility of recurrence when the treatment stops<sup>23</sup>. Moreover, abdominal wall or scar endometriosis is less responsive to the hormonal treatment. Surgical resection on healthy tissue is the treatment of choice. To achieve healthy tissues may necessitate partial resection of the deep fascia<sup>24</sup>. In patients with wide gaps in the abdominal wall the use of a mesh is suggested<sup>1</sup>. The surgical treatment is successful in 95% of the cases, while the recurrence percentage is 4.3%<sup>25</sup>. The local recurrence could be generated from a non-sufficient resection of the endometrium focus. Thus the patients should have a postoperative follow up because of the possibility for recurrence. Wide excision, with at least 1cm margin, is considered the treatment if choice, even for recurrent lesion<sup>26</sup>. The combination of a surgical re-excision and hormonal treatment is also recommended<sup>23</sup>. The prevention of scar endometriosis may also be important. During gynecological and obstetrical operations, there should be a careful hemostasis and flushing of the abdomen and of the incision before the final closure<sup>27</sup>. A different needle should also be used for the closure of the abdominal wall than the one used for the uterus. Surgical tools and gloves should be changed. A wound edge protector should be used to separate the incision from the peritoneal cavity. This protective cover prevents seeding of endometrial cells into the incision<sup>28</sup>.

## Conclusion

Abdominal wall endometriosis is found in women of reproductive age that have formerly undergone gynecological or obstetrical operations. It is currently regarded as a rare pathological condition, however it may be more frequent than generally assumed, due to the increase of Cesarean section rate. Because the symptoms are not always typical, high clinical suspicion is needed for the right diagnosis not only by the physicians in the field of obstetrics and gynecology but also by general surgeons. The surgical treatment is the treatment of choice and postoperative follow up is necessary. In conclusion, the impor-

tance of these cases is to emphasize that scar endometriosis should be on the differential diagnosis for females complaining of mass or pain in or around the scar after gynecological procedures and surgical resection should be offered to nearly every patient with excellent results.

## Riassunto

**INTRODUZIONE:** L'endometriosi è una condizione patologica ginecologica caratterizzata dall'impianto di tessuto endometriale funzionante al di fuori della cavità uterina. Riguarda principalmente le donne in età riproduttiva, con una incidenza a livello stimata attorno al 8-15%. Il tessuto endometriale ectopico può essere trovato in localizzazioni intrapelviche ed extrapelviche. Un localizzazione rara dell'endometriosi è l'impianto su incisione chirurgica, e più comunemente la zona di incisione del taglio cesareo o dell'isterectomia. L'endometriosi su cicatrice chirurgica spesso non viene diagnosticata correttamente poiché simula molte condizioni chirurgiche tra cui il laparocele o i lipomi. La diagnosi è basata principalmente su un alto grado di sospetto.

**OBIETTIVO:** Lo scopo di questo studio è di evidenziare le caratteristiche di questa rara patologia e di valutare l'algoritmo diagnostico e le opzioni terapeutiche.

**METODO/MATERIALE:** Nel presente studio presentiamo una serie di sette casi riscontrati nel nostro dipartimento tra il 2014 ed il 2016, oltre ad una revisione della letteratura internazionale per la diagnosi e il trattamento dell'endometriosi su incisione chirurgica.

**CONCLUSIONI:** L'eziologia della malattia non è completamente nota e si sono sviluppate diverse teorie. I sintomi non sono tipici in questa patologia ed è spesso una scoperta postoperatoria. Il sintomo più tipico dell'endometriosi su incisione chirurgica è il dolore locale periodico in combinazione con una massa palpabile localmente che di solito si verifica nel periodo mediano del ciclo mestruale. La diagnosi differenziale può essere posta con varie condizioni e la diagnosi si basa principalmente su un elevato sospetto clinico. L'uso di strumenti, come l'ecografia, il color Doppler, la TC e MRI, sono utili per migliorare la diagnosi.

Vengono suggeriti gli elementi per la prevenzione dell'endometriosi post-chirurgica.

Il trattamento di scelta è la rimozione chirurgica delle lesioni con margini liberi, che è seguita dalla guarigione in circa il 95% dei casi. In alternativa, può essere adottato un trattamento farmacologico con farmaci contraccettivi orali combinati, ma la risposta è per lo più parziale ed è alto il tasso di recidiva.

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