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Endometriosis is seen in women during their reproductive age, where functional endometrial glands of the uterus and stromal component are observed outside the uterine cavity. Endometriosis in an operative scar is a rare event following mainly obstetric and gynecologic operation. Typical signs of scar endometriosis are cyclic pain and swelling tumor in the scar after obstetric or gynecologic operations. We present 24 cases of scar endometriosis with discussion and emphasis on variants of clinical signs, differential diagnostic, methods of treatment, and prevention.

Keywords: endometriosis, scar, laparotomy, incision, cesarean section

1. Introduction

Endometriosis is one of the most common diseases of the female genital tract. This pathology is characterized by the presence of the endometrial tissue outside the place of its usual localization. Commonly, in gynecological practice, we have to face pelvic endometriosis; however, extragenital endometriosis is described in different organs and body systems, including the intestines, urinary tract, thorax, umbilicus, etc. It is very difficult to establish the real frequency of extragenital endometriosis [1]. This is due to different approaches to accounting of the disease, treatment of patients in a variety of clinics, and underestimating of non-severe clinical signs. Among locations of endometriosis in the urinary tract, the lesion of the bladder is dominant, and in the gastrointestinal tract, lesions of the rectum and sigmoid colon [1]. Affection of the lungs occurs significantly rare. Endometriosis in a postoperative scar is a secondary process in scars after surgical procedures affecting the endometrium: cesarean section, hysterectomy, amniocentesis, etc. Nevertheless scar endometriosis occurs also after general surgery—appendectomy, cholecystectomy, and correction of hernias [2]. The term “endometrioma” is used for well-marked tumor-like lesions [3]. However, there may be a situation characterized by a typical clinic of endometriosis in the absence of a clearly defined lesion, and this makes diagnosis difficult. Its clinical diagnosis is confused with abscess, hematoma, suture granuloma, desmoid tumor, sarcoma, etc. Incidence rates for endometrioma associated with cesarean section have been reported to be 0.01–4% [3]. The incidence of endometrioma in episiotomy scars is much less than in abdominal wall scars. In this study we present 24 cases of endometriomas appearing after cesarean section, laparoscopic cystectomy, and perineal incision.
2. Case reports and discussion

We present 24 cases of scar endometriosis that we observed since 2003 till 2018. Mean age of patients was 29 years, in the range of 25–33.5 years. Twenty-two patients have previously undergone cesarean section, five of them were operated twice. Cesarean section was performed 5 (range 4–7) years ago. Clinical signs appeared during 1–3 years after the last cesarean section. More than half of the patients initially seek care from general surgeons and only after the examination were sent to a gynecologist. One patient suffers from endometriosis of postoperative scar after perineorrhaphy performed 23 years ago, and the last patient presents lesion in the region of left lateral trocar tract after removal of endometrioid ovarian cyst 6 months ago. It is interesting that 12 patients believed that the cause of their suffering was a non-gynecological disease, so they initially claimed to a general surgeon.

All patients after previous cesarean section presented painful tumor of a scar; palpable sizes of lesion were 45 (range 35–55) mm. Sometimes the lesion rises above the skin, but usually it is palpable deep in the tissue. The degree of pain increased during menses, and tumors became more swollen at that time—but only in 50% cases. Another 50% of patients suffered only from the presence of swelling in the area of the postoperative scar with moderate pain that was not associated with the menstrual cycle. One patient presented fistula in the angle of a scar with dark brown discharges during menstruation (Figure 1).

Endometriosis in the region of the perineum developed in a patient after childbirth for 14 years. She obtained medical care only after a slowly increasing lesion began to cause discomfort during sexual activity and walking. It was an unmovable swelling, 5–4–4 cm in sizes, almost woody in consistency. The moderate pain syndrome was not of a cyclical nature. Initially we suspected the pathology of Bartholin's gland, but later we were inclined to think about postoperative scar endometriosis. It should be noted that none of the patients examined by us had a history of genital endometriosis, although according to the literature this may be one in every fourth case [1].

All patients have undergone physical examination with obligatory ultrasound of postoperative scar for visualization of endometrioma (Figure 2). On ultrasound examination in all cases, we revealed hypoechoic nodules with irregular borders (Figure 3). Palpable tumors may have oval or irregular shape with or without clear marked boundaries. Internal structure was homogeneous only in one case; in others, tumors demonstrated different combinations of low echogenicity, hyperechoic...
inclusions, and anechoic cavities. All lesions were determined by good vascularization. It should be noted that in almost all cases the size of the structures on ultrasound was significantly less than palpable. It can be explained by the presence of perifocal inflammation.

All patients have undergone excision of tumors under general anesthesia with subsequent histological examination of the removed tissues (Figure 4). In 19 cases lesions were bordered by aponeurosis, muscles were intact, and wounds were sutured in layers.

Macroscopically excised structures usually were presented as a dense-consistency tissue with well-marked brown spots after incision (Figure 5) or without it (Figure 6). In three cases after operation, seroma with subfebrile temperature was formed; it was treated with antibiotics, aspiration drainage, and compressive bandage. Lesions were healed by secondary intension.
The last three cases were not ordinary because lesions extended down to the subperitoneal layer and even to the peritoneal cavity. Localization of tumor in subperitoneal space was confirmed by MRI because the palpation data and the ultrasound results were doubtful in spite of typical “endometrioid” complain (Figure 7).

The patient suffered also from bilateral ovarian cysts (not endometrioid!) and has undergone laparoscopic cystectomy. And during the operation, we could visualize an endometrioid tumor located subperitoneally (Figure 8).

In two cases we have to perform laparotomy after excision of endometrioid lesion due to its spread to the abdominal cavity. In the first case, we removed tumor, observed pelvic cavity, found no signs of pelvic endometriosis, and repaired surgical wound. In the second case, tumor also spread to the abdominal cavity and connected with the low segment of the uterus. During excision we diagnosed injury of the posterior wall of the urinary bladder. The trauma of the urinary bladder was sutured by a urologist, Foley’s catheter was inserted for 8 days, and wound was repaired. In this case we also revealed no evidence of pelvic endometriosis [4].

Excision of mass after perineorrhaphy was technically difficult due to woody consistency and severe adhesions with surrounding tissues.

Histological examination of the removed tissues revealed the presence of endometriosis in all cases except one. Endometrial tissue was in various proportions.
Endometriosis of Postoperative Scar
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with fibrous and fat tissues, even with suppuration of endometrial structures (after excision of lesion in the perineal region).

In all the cases, no recurrence was detected during the follow-up (1–12 years) except one—but it should not be considered as a recurrence, as the lesion appeared on the other side of the postoperative scar.

The final case was more interesting. As mentioned before, a 50-year-old patient was operated 6 months ago—bilateral adnexectomy by laparoscopic approach was performed due to endometrioid ovary cysts from both sides. Five months later the patient found the swelling painful mass in the region of the left trocar tract. Pain gradually increased, and slight redness of the skin appeared above the palpable tumor (Figure 9).

Adenocarcinoma was defined by histological examination after excision of tumor. We suppose that adenocarcinoma probably took place at the moment of the first laparoscopic operation in some parts of a cyst but was not revealed, so it was possible to develop into malignant tumor in trocar wound (we found out that the
extraction of a cyst from the abdominal cavity was performed without using special containers). From the other side, it is difficult to exclude endometrial transformation in a malignant tumor [5]. Later the patient underwent several courses of chemotherapy.

Some theories have been put forward to explain the pathogenesis of endometriosis, such as lymphatic or hematogenous dissemination, coelomic metaplasia, and cell immunity change theory. The most prominent theory is that reflux of endometrial cells through the tubes into the peritoneal cavity during menstruation leads to pelvic endometriosis [2]. The viability and growth potential of desquamated menstrual endometrium have been demonstrated [6]. The metaplasia theory states that endometrioma arises due to metaplasia of pluripotential mesenchymal cells [3]. The transport theory suggests that endometrial cells may be transported to distant location, forming endometriomas during surgical procedures. The cause of surgical scar endometriosis is believed to be iatrogenic transplantation of the endometrium to the surgical wound. Evolution of knowledge about scar endometriosis is rather interesting. Review of the early literature shows a high association of abdominal scar endometriosis with a previous ventrofixation operation [7]. Then the operation of hysterotomy for termination of early pregnancy has been increasingly associated with this condition [6], and it was shown that early pregnancy endometrium was easier to transplant than the term pregnancy endometrium. In our study, one patient also was after termination of pregnancy by cesarean section at 18 weeks.
due to severe portal hypertension. In recent years, both genetic predisposition to endometriosis and the role of epigenetic factors are also actively discussed [8].

Nowadays, increasing of cesarean delivery—the main reason of abdominal scar endometriosis. Anyway, any operation on the uterus—it does not matter either ventrofixation or cesarean section—may lead to scar endometriosis due to the phenomenal viability of the endometrium [6, 7].

Endometriosis of postoperative scar can develop both after surgery performed by laparotomic approach and after laparoscopy in the area of the trocar opening, as well as in the perineal region after its incision during childbirth. Regardless of the type of intervention, the crucial point is that the endometrium implants on the wound surface and develops after it. Therefore, postoperative scar endometriosis is an example of iatrogenic endometrial transplantation to the wound surface. It is the transplantation theory which explains the appearance of the endometrium in the area of postoperative scar after non-gynecological operations—appendectomy, cholecystectomy, etc. In these cases, the operation is performed either during or immediately after menstruation, when the presence of the endometrium in the abdominal cavity is possibly as a result of reflux of endometrial cells through the fallopian tubes.

Despite the ectopic location, endometrial tissue is able to respond to hormonal effects, thereby causing clinical signs of disease. Endometriosis of postoperative scar is a typical example of extragenital endometriosis. But endometriosis of the postoperative scar is not only damage to the skin. In this situation, it is necessary to discuss the lesion of all tissues that were affected during surgical interventions.

The most common presenting symptom of endometrioma in a scar is a palpable mass associated with cyclic pain and swelling during menses [2]. Sometimes endometriomas may be multiple. Endometrial implants behave like normal endometrium in their response to hormones. Ovarian hormonal action on ectopic endometrial cells during menstrual period causes slight bleeding at the scar location with an inflammatory reaction and subsequent tissue repair. Thus, as each menstrual cycle goes by, the lesion increases in sizes, and this increasing might compromise the skin, subcutaneous cellular tissue, aponeurosis, and peritoneum. We observed these events in three of 19 cases. Also we consider that inflammatory changes of endometrial mass resulted in injuring the urinary bladder during excision.

If the symptoms are cyclic in the woman with a prior history of surgery on the uterus, then endometriosis should be the most likely consideration. It is practically pathognomonic. There is no need for advanced propaedeutics, and the diagnosis may be based on anamnesis and physical examination. Some authors describe a characteristic triad of periodic pain, tumor, and history of cesarean section [9]. When the patient complains are not cyclical, clinical diagnosis is impaired. Noncyclical symptoms are observed in 25–45% of patients with scar endometriosis. An association between scar endometriosis and pelvic endometriosis is possible to find in one quarter of the cases [9], but in our study we found none. The differential diagnosis of a mass in a scar includes keloid formation, suture granuloma (Figure 10), hematomas, abscess, desmoid tumor, postoperative hernia, lipoma, cyst, or even strange body.

It is also possible to face with a rare condition—the so-called gossipiboma. It is a foreign body-related inflammatory pseudotumor caused by retained non-resorbable or partially resorbable substances [10]. We observed a patient with cutaneous fistula, as casuistically it is possible to give an example of unusual clinical situation of a patient with an endometriotic uterocutaneous fistula. The patient presented a painful nodule on the cesarean scar, which was bleeding during menstruation.
It was established that the lesion extended to the uterine fundus, connecting the endometrial cavity with the skin [11]. Ultrasound examination and MRI may sometime aid in the diagnosis of scar mass, and fine needle aspiration cytology also has certain value in the diagnosis of scar endometriomas. The typical sonographic pattern is the presence of subcutaneous nodule, hypoechoic with hyperechoic strands and irregular margins. Sometime it is possible to visualize complete or incomplete hyperechoic ring around the nodule caused by a perifocal inflammatory reaction. In lesions larger than 3 cm, small cystic areas may be detected, possibly because of recent hemorrhage. In the typical case, a single peripheral vascular pedicle with arterial flow entering the nodule can be shown by color Doppler investigation. In very small lesions, this sign may be absent [12]. Because of high resolution of MRI, this technique makes it possible to identify smaller lesions and distinguish signs of organized hemorrhages within endometriomas. Moreover, MRI has better performance than computed tomography scans in relation to outlining the subcutaneous, muscle, and aponeurotic tissue layers (Figure 7).

In some cases thin needle puncture guided by ultrasound with cytological analysis helps to confirm diagnosis. In our study five of 24 patients have undergone this procedure before incision. However, its use is still controversial because of the risk of causing new implants at the puncture sites or perforating a hollow organ in the case of unrevealed or incarcerated hernia that simulated endometrioma.

Anyway imaging modalities remain nonspecific and do not modify the plan of treatment—wide surgical excision. Therapy with oral contraceptives, progestins, medroxyprogesterone acetate, and gonadotropin-releasing hormone agonists has been tried with minimal effects [4, 8]. In some patients the effects can be relatively long-lasting, but complete, permanent regression of endometriosis is rare with medical therapy. In our study we observed a patient treated with levonorgestrel-releasing intrauterine device during 1.5 years. Clinical signs of endometriosis disappeared, but lesion in the region of postoperative scar remained. After excision of this tumor, we found no typical “chocolate” inside (Figure 11).

Histological examination revealed “hollow” glands without epithelial cells. Treatment of this patient resulted in inhibition of endometrial tissue, but stromal component of endometriosis remained.

That is why for endometriosis of postoperative scar, total surgical excision is considered to be gold standard for both diagnosis and treatment. Resection must be complete with clear margins to prevent recurrence. The excision may be technically difficult depending on the depth and the size of mass. It is possible to use coagulation; it leads to smaller bleeding from infiltrated surrounded tissues. Sometimes
large defects in aponeurosis after excision require polypropylene mesh for repair-
ing. Surgery should be performed some days before the menstrual period in order to
avoid an inflammatory reaction and make tissue removal easier.

Scar endometriosis as well as endometriosis at other sites can become malignant.
It is a rare event occurring in 0.3–1% of scar endometriomas. The phenomenon of
malignancy arising in association with endometriosis was first described in 1925.
Clear cell carcinoma is the most common histological subtype [5]. Frequent recur-
rence might indicate malignant degeneration of tumor. That is why longtime clini-
cal follow-up is strongly recommended because malignant transformation might
vary from a few months to more than 40 years.

In order to prevent scar endometriosis, some measures have been proposed.
First of all, it is reasonable to close the peritoneal and visceral peritoneum with
sutures at the time of cesarean section and perform an introflexed suture of the
uterine incision and parietal peritoneum. Refusing these measures may increase
the postoperative occurrence of an endometrioma in the scar. Second, it is not
recommended to elevate the uterus out of the abdominal cavity during cesarean
section or hysterotomy. Also it is recommended not to use the same instruments
for hysterorrhaphy and suturing abdominal wall layers. At last, at the end of the
surgery, the abdominal wall wound should be cleaned thoroughly and irrigated
with high solution before closure. No measures of prevention have proven its
efficiency, and all these measures were suggested without any evident scientific
corroboration.

3. Conclusions

Twenty-four cases of endometriosis of postoperative scar have been presented.
The occurrence of this type of extrapelvic endometriosis is supported by the iatro-
genic implantation theory. Cyclic pain and swelling tumor in the scar after obstetric
or gynecologic operation are typical signs of scar endometriosis. The absence of
cyclic pain syndrome in the presence of a lesion in the region of the scar requires
a differential diagnosis. Wide excision of tumor is the best way for treatment and
final diagnosis.

Conflict of interest

The author declares no conflict of interest.
References


