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Case Report / Приказ болесника

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Abdominal wall endometriosis: clinical presentation, imaging features and management of five cases

Ендометриоза предњег трбушног зида: клиничка презентација, имиџинг карактеристике и начини решавања – приказ пет случајева

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SUMMARY

Introduction Endometriosis represents a functional endometrium outside the uterine cavity. Ectopic endometrial tissue has been identified within the pelvis, affecting both pelvic and extrapelvic organs causing recurrent pelvic or abdominal pain corresponding to the menstrual cycle.

The incidence of abdominal wall endometriosis after cesarean section is approximately 2%. It is often mistaken for other conditions primarily because this condition is underestimated on imaging.

The objective of this case series is to summarize possible clinical and radiological presentations of this uncommon condition.

Outline of cases The authors present a case series of five patients who developed abdominal wall endometriosis after the Cesarean section. Having been diagnosed clinically, the patients underwent open abdominal surgery, and were treated by surgical resection.

Conclusion Good clinical practice and excellent surgical techniques may help in preventing endometriosis, while adequate clinical examination and proper imaging can help in presurgical planning and successful definitive treatment.

Keywords: abdominal wall; caesarean section; endometriosis; endometrioma; imaging; surgery

САЖЕТАК

Увод Ендометриоза представља функционални ендометријум изван материчне дупље. Ектопично ткиво ендометријума је идентификовано унутар карлице, захвата карличне и екстрапелвичне органе узрокујући пелвични или бол предњег трбушног зида који је синхрон циклалном менструалном крварењу.

Учесталост ендометриозе предњег трбушног зида након царског реза је 2% и често се посумња на друга патолошка стања првенствено услед непрепознавања овог стања на доступним имицинг модалитетима.

Циљ ове серије случајева је приказати могуће клиничке и радиолошке презентације ендометриозе.

Приказ болесника Приказана је група од пет пацијенткиња са ендометриозом предњег трбушног зида, након царског реза, која је клинички дијагностикована и подвргнута отвореној хирургији локалном ексцизијом.

Закључак Добра клиничка пракса и извршне хируршке технике могу помоћи у спречавању ендометриозе, док адекватан клинички преглед и одговарајући имицинг могу помоћи у хируршком планирању и успешном коначном лечењу.

Кључне речи: трбушни зид; царски рез; ендометриоза; ендометриом; имицинг дијагностика; операција

INTRODUCTION

Abdominal wall endometriosis (AWE) represents a challenging and underestimated diagnosis for both clinicians and radiologists due to its rarity [1-3].

However, it seems to be the most frequent localization of the extra-pelvic endometriosis [3, 4]. In the past, imaging did not represent a significant part of presurgical assessment, providing only scarce information on general radiological features of AWE [3].

Endometriosis is reported in up to 10% of menstruating women and in up to 40% of reproductive age women who undergo gynecological procedures allowing endometrial tissue to be transplanted [5].

Anterior wall endometriosis should be taken into consideration when a painful and tender palpable abdominal wall mass appears within or near cesarean section, hysterectomy, or laparoscopic trocar sites, with or without bleeding from Cesarean section scar and cramp [6].

Presurgical assessment of this condition should consist primarily of clinical examination followed by appropriate imaging, most commonly ultrasonography (US) or magnetic resonance imaging (MRI).

The aim of this case series is to summarize possible clinical and radiological presentations of this uncommon condition.

CASE REPORTS

Case 1

A 33-year-old patient was referred to a gynecologist due to pain in the middle of Cesarean section scar that correlates with menstrual bleeding. The patient underwent two Cesarean sections 18 and 49 months before.

Clinical examination revealed two palpable tumors, being approximately 25x25mm and 30x20mm in size in the abdominal wall just beneath the Cesarean section scar, that being in accordance with US findings of two irregular, heterogeneous subcutaneous nodules of 23x21mm and 32x23mm. MRI confirmed diagnosis, showing two spiculated masses within the muscle of the anterior abdominal wall with imprints to the peritoneum.

Case 2

A 37-year-old patient was referred to a gynecologist due to pain in the left part of Cesarean section scar correlating with menstrual bleeding. The patient was delivered via Cesarean section 25 months before.

Clinical examination revealed a palpable tumor, measuring approximately 45x30mm, in the abdominal wall just beneath the healthy Cesarean section scar, and it correlated with US findings of irregular, heterogeneous subcutaneous nodules of 50x30mm with internal scattered echoes typical for scar endometriosis.

Case 3

A 32-year-old patient was referred to a gynecologist due to pain in the right part of Cesarean section scar that correlated with menstrual bleeding. The patient had a history of one previous Cesarean section 31 months before.

Clinical examination revealed a palpable tumor, whose approximate size was 25x15mm, in the abdominal wall just beneath the Cesarean section scar. CT confirmed diagnosis, showing one spiculated hypodense mass lesion adjacent to the scar.

Case 4

A 31 year-old patient suffered from a pain in the middle and right part of Cesarean section scar and she was therefore referred to a gynecologist. She had undergone Cesarean section 13 months before .

Clinical examination revealed a palpable tumor, measuring approximately 35x20mm, in the abdominal wall just beneath the Cesarean section scar, which correlated with US findings of irregular, subcutaneous nodule of 37x18mm.

Case 5

A 41-year-old patient was referred to a gynecologist because she had complained of the pain in the left part of Cesarean section scar with cutaneous bleeding fistula that correlated with menstrual bleeding. The patient underwent two Cesarean sections, 94 and 28 months before.

Clinical examination revealed a palpable tumor, its approximate size being 30x25mm, in the abdominal wall just beneath the Cesarean section scar, which was in accordance with US findings of irregular, heterogeneous subcutaneous nodule of 33x27mm. MRI confirmed this diagnosis showing spiculated mass within the muscle of the anterior abdominal wall with imprints to peritoneum and associated with cutaneous fistula.

All five patients underwent mini-laparotomy with surgical approach via previous Cesarean section scar. In all cases, endometriomas were completely surgically removed. The abdominal wall defect was reconstructed by sutures placed through the anatomical layers of abdominal wall (Table 1.).

Being a part of the abdominal wall, all surgical specimens were pink-brown in color. However, after the resection of specimens, they became multicoloured in appearance and of elastic consistency (Figure 2, Figure 3).

Pathohistological examinations of collected samples revealed a lot of coalescent foci of endometrial mucosa consisting of dilated endometrial glands, fenced by stromal cells with typical morphological characteristics. Signs of fresh and old hemorrhage were detected in examined materials (Figure 4a).

Progressive destruction of the anterior abdominal wall muscles was due to anastomosed stripes of newly developed fibrous tissue (Figure 4b).

The presence of all elements of the endometrial lining within the anterior abdominal wall as an ectopic location confirmed the PH diagnosis.

DISCUSSION

Anterior abdominal wall endometriosis is closely connected with Cesarean section, its reported incidence going up to 2% of all women who underwent cesarean delivery [5, 7, 8].

Conditions such as stitch granuloma, hematoma, keloids, primary or metastatic adenocarcinoma and incisional hernia create a diagnostic dilemma [5, 8].

The time period from Cesarean section to the onset of symptoms ranges from a month to 17.5 years, the average being 30 months [8, 9].

Direct implantation is the most exploited theory of pathogenesis of abdominal wall endometriosis. Namely, during the surgical procedure that consists of opening the uterine cavity, endometrial tissue is implanted into the surgical scar and afterwards proliferates following the same hormonal influences as uterine endometrium [9]. Endometriosis of surgical scar often infiltrates not only superficial layers of the abdominal wall, but also deeper layers - including rectus muscle and peritoneum [8].

A common clinical symptom of AWE is cyclical pain spreading within the abdominal wall, usually occurring at the time of menstruation [10].

Palpable mass at the site of maximum tenderness in the region of the surgical scar, which is sometimes multiloculated, is a typical clinical finding [8,10].

Anterior abdominal wall endometriosis can also be asymptomatic and incidentally discovered during imaging performed for other reasons [8]. Routine imaging modality for the assessment of AWE is ultrasound (US), due to its availability and low cost. Endometriomas are usually found in the midline and in the left hemiabdomen. Echosonographic features of scar endometriomas are various and non-specific [2]. Most commonly, they present as solid, nodular masses, sometimes stellate clearly defined margins [1]. Recent research has confirmed that heterogenous echogenicity is most common, representing the distribution of fibrous and hemorrhagic components, and varying across the menstrual cycle [1,3]. Internal echogenic spots or thick strands represent fibrotic tissue component [2]. On power Doppler examinations, the lesions show internal vascularity, peripheral in distribution. It is important to notice that US findings of scar endometriomas differ from adnexal ones, typically

presenting with round cystic masses, clear margins, thickened walls, and low-level internal echoes [2].

The main role of computed tomography (CT) and magnetic resonance imaging (MRI) is to depict the exact extent of the disease preoperatively. CT may be performed with or without use of contrast agent - contrast studies are preferable due to higher sensitivity and specificity [2]. The CT presentation of AWE nodules is usually non-specific: solid soft-tissue masses that are of heterogenous density and directly associated with the scar tissue. Contrast enhancement of the lesions is mild to moderate, with depiction of the feeding vessels within or adjacent to the nodule [2]. Internally, it can contain blood products with variable attenuation (dependent on chronicity and menstrual cycle phase) [3].

MRI is a preferred cross-sectional imaging method because of the best tissue characterization, lack of ionizing radiation and possibility of evaluating the relationship between scar tissue and AWE, mandatory for presurgical planning. Endometrioma is usually T2 hyperintense heterogenous nodule with hemorrhagic foci (best observed on T1), directly associated to scar tissue. On T1 sequences with fat suppression, endometrial foci are seen as heterogenous hyperintense lesions, compared to the abdominal musculature, as a result of hemorrhage. Variable T2 signal intensity is associated with cyclic hormonal influences, chronic inflammation and fibrosis [11]. Contrast enhancement is usually strong, at least in a portion of AWE. Diffusion-weighted imaging is used for diagnosing this condition, but apparent diffusion coefficient (ADC) values also vary across the menstrual cycle [12].

Diagnostic rates on CT and US are low, with undiagnosed endometriosis in 77-100% of the cases, while MRI establishes the correct diagnosis in 75% of the cases [3]. It seems that radiologists are in general not familiar with the spectrum of endometriosis imaging features. The most common differential diagnoses include: dermoid tumors, hematomas, keloids, granulomas, metastases and lymphoma [13,14]. Dermoid tumors may mimic the radiological appearance of endometriomas but lack the cyclic changes. Hematomas are associated to a recent trauma, they lack typical contrast enhancement and are of a sudden onset. Keloids are usually indistinguishable from AWEs and require biopsy [15].

With emerging of new, less invasive therapeutic approaches, such as high-intensity focused US (HIFU) and radiofrequency ablation (RFA), the requirements for the adequate

imaging follow-up will raise, and so should the radiological awareness of this condition and its variable presentation on imaging [16].

Only the presence of endometrial glands and stroma within the lesion can confirm histopathological diagnosis of endometriosis [17].

Therapeutic options for abdominal wall endometriosis include surgical excision and medical therapy with hormonal agents -progestagens, danazol, oral contraceptive pills and gonadotropin-releasing hormone (GnRH) analogs but with a partial response and recurrence when medications are discontinued. The only definitive treatment is a wide local surgical excision with clear margins to prevent local recurrence [8, 18, 19, 20].

CONCLUSION

Anterior abdominal wall scar endometriosis after a cesarean section represents a challenge from both diagnostic and therapeutic aspect.

With a rising Cesarean section rate, anterior abdominal wall scar endometriosis is becoming more common, considering the theory of iatrogenic endometrial inoculation into the surgical wound.

Good clinical practice and excellent surgical techniques may help in preventing endometriosis, while adequate clinical examination and proper imaging can help in presurgical planning and successful definitive treatment.

DISCLOSURE

Conflict of interest: None declared.

Ethical compliance statement:

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

Ethical standards:

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Written consent to publish all shown material was obtained from the patient.

Paper accepted

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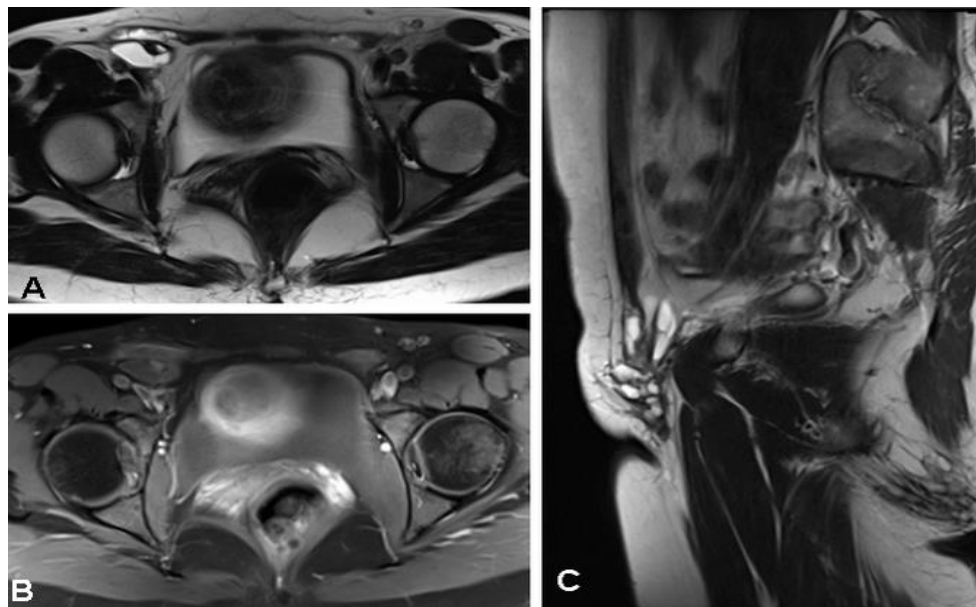


Figure 1. Abdominal wall endometriosis; multicompartmental cystic lesion is observed at the level of right inguinum on T2W axial image (A), with slight contrast enhancement of the walls seen on T1W axial image (B), extending craniocaudally below the level of anterior abdominal wall (C – T2W coronal image)



Figure 2. Anterior abdominal wall fragment with the zones of hemorrhage and fibrosis

Paper accepted



Figure 3. Resected abdominal wall with cutaneous fistula

Paper accepted

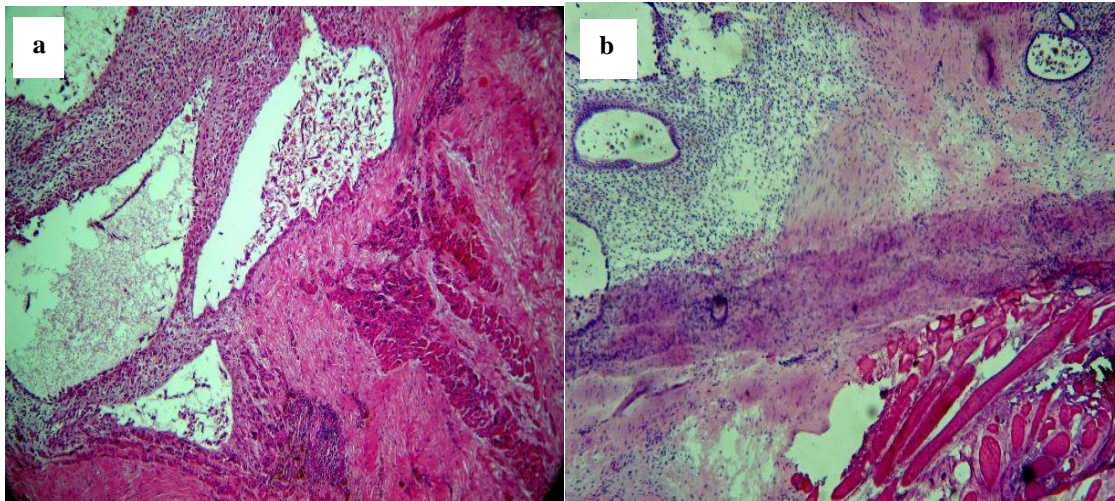


Figure 4. a) Confluent endometriosis foci in the abdominal wall tissue (H&E, 10 × 10); b) fibrous destruction of the muscular beam induced by confluent endometriosis zones (H&E, 10 × 25)

Table 1. Clinical information of all five cases

Patient No.	Age at diagnosis	Months after CS	Main symptom	Impression after physical examination	Imaging used for diagnosis	Year of operation for AWEC
1	33	49, 18	Painful mass in ACSS relationship with menstruation	Endometrioma	USG, MRI	2015
2	37	25	Painful mass in ACSS relationship with menstruation	Endometrioma	USG	2017
3	32	31	Painful mass in ACSS relationship with menstruation	Endometrioma	CT	2018
4	31	13	Painful mass in ACSS	Granuloma	USG	2018
5	41	94, 28	Painful mass in ACSS with cutaneous fistula relationship with menstruation	Endometrioma	USG, MRI	2019

CS – Cesarean section; ACSS – abdominal wall previous Cesarean section scar; AWEC – abdominal wall endometriosis in CS scar; MRI – magnetic resonance imaging; CT – computed tomography; USG – ultrasonography