Parietal Endometriosis: A Challenge for the General Surgeon

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Rezumat

Endometrioza parietală: o provocare pentru chirurgul generalist

Introducere: Endometrioza parietală (EP) este o patologie rară, care apare frecvent la femeile fertile, după intervenții chirurgicale sau ginecologice. Simptomatologia cvasipatognomonică constă în durere catamenială asociind sau nu formațiune palpabilă. Diagnosticul ridică probleme, deoarece adesea se poate confunda cu granulomul de fir, hematomul, hernia și cancerul.

Pacienți și metode: Între ianuarie 2007 și decembrie 2017, 10 pacienți cu EP au fost internate în clinica noastră pentru diagnostic și tratament.

Rezultate: Vârsta medie a pacientelor a fost de 35,8 ani. Simptomul principal a fost durerea abdominală (9/10 pacienți) și examenul clinic a relevat prezența unei formațiuni palpabile la toate pacientele. Cinci pacienți au fost diagnosticate preoperator cu EP, iar cinci incorect diagnosticate cu tumoră de perete abdominal (4 pacienți) și granulom de fir (1 pacientă). Opt pacienți au avut istoric de intervenții ginecologice (operație cezariană, epiziotomie), iar 2 pacienți nu au avut operații în antecedente. Dimensiunile tumorilor au fost de 1-14 cm. Rezecția tumorală a necesitat reconstrucția peretelui abdominal cu plasă de întărire sau substituție la 5 pacienți, iar la restul plasa nu a fost necesară.

Concluzii: Studiul nostru confirmă că EP reprezintă o entitate chirurgicală rară și sugerează necesitatea anamnezei și examenului

Received: 26.02.2018

Accepted: 10.04.2018

clinic minuțios, dar și efectuarea examenelor imagistice, pentru elucidarea diagnosticului corect, în vederea stabilirii tratamentului chirurgical adecvat.

Cuvinte cheie: endometrioza parietală, endometrioza perineală, endometrioza abdominală, endometrioza cicatriceală, endometriom

Abstract

Background: Parietal endometriosis (PE) is a rare pathology, which usually develops in fertile women, after surgical or gynecological procedures. Its quasi-pathognomonic symptomatology consists in catamenial pain with or without palpable mass. The diagnosis can be challenging because it may be confused with stitch granuloma, hematoma, hernia or even cancer.

Patients and methods: Between January 2007 and December 2017, 10 female patients with PE were referred for diagnosis and surgery to our clinic.

Results: The mean age of the patients was 35.8 years. The primary symptom was pain (9/10 patients) and a palpable mass was present in all patients. Five cases were correctly preoperatively diagnosed as PE and five were misdiagnosed as tumors (4 patients) and stitch granuloma (1 patient). Eight patients had a history of gynecological procedure (cesarean section, episiotomy) and two had no previous surgical interventions. The size of the mass varied from 1 cm to 14 cm. Resection of PE required parietal reconstruction with mesh in five patients but for the rest of the patients no mesh was needed.

Conclusions: Our study confirms PE as a rare surgical entity and indicates the necessity of thorough history and physical examination, as well as imaging exams, for making the correct diagnosis in order to choose the appropriate surgical procedure.

Key words: parietal endometriosis, perineal endometriosis, abdominal endometriosis, scar endometriosis, endometrioma

Background

Although Karl Freiherr von Rokitansky was believed to describe in 1860 the first case of endometrioma, referring to the disease as “sarcoma”, the first unequivocal report of the presence of endometrial tissue within an ovary is contained in a paper published in 1899 by Russel, who incidentally found in the left ovary of a woman some ‘areas, which were an exact prototype of the uterine glands and interglandular connective tissue’ (cited by 1). Therefore, endometriosis is defined as the extrauterine spread of functional endometrial glands and stroma. It occurs in up to 15% of women of reproductive age (2). The most commonly affected organs are pouch of Douglas,

ovaries and peritoneum covering the pelvic organs, followed by the bowel and urinary tract. Involvement of extrapelvic sites (e.g. central nervous system, lungs, pleura, kidney, lymph nodes, extremities, umbilicus, hernia sacs, bronchi, gallbladder, bladder, small intestine, large intestine, appendix, omentum, nasolacrimal canal) is unusual (3-7) as well as abdominal wall involvement. Abdominal wall endometriosis is defined as the presence of endometrial tissue superficial to peritoneum (8). Parietal endometriosis (PE) is a definition that includes not only the abdominal wall but also the perineal endometriosis. PE can be easily confused with an abscess, lipoma, hematoma, sebaceous cyst, stitch granuloma, incisional hernia or tumors (9), that can delay

the surgical treatment or be followed by inappropriate surgical procedures, causing emotional and physical distress of the patient (10). The aim of this study is to report the experience of a Romanian general surgery center in diagnosing and treatment of this rare entity.

Materials and Methods

A retrospective, observational study with a case series of patients from the 130-bed Department of General Surgery and Liver Transplantation, Fundeni Clinical Hospital, was conducted from January 2007 to December 2017. Patients with histopathologically confirmed PE were

identified from the surgical database of our institution. A total of 10 patients with PE were found in department's medical records. Data regarding age, signs and symptoms, present and past history of disorder, family pathology, imagistic findings, type of surgical intervention and postoperative course were analyzed.

Results

The highest addressability in our study was found to be in 2015 (3 cases) (*Table 1*). The study group is comprised of 10 women with mean age of 35.8 years (with range from 24 to 74 years). Five patients presented with abdominal mass in (*Fig. 1*) or above (*Fig. 2*) the

Table 1. Characteristics of the patients with PE

No.	Year of address.	Age (years)	Chief complaint	Surgical history	Initial diagnosis	Localization	Maximal diameter	Imaging study	Hospitalization (days)	Surgery
1	2007	32	cyclic pain	none	tumor	inguinal	5 cm	US	6	EM
2	2007	32	mass with d.v.	none	tumor	inguinal	1.5 cm	US, IRM	2	E
3	2010	74	pain	C/S	tumor	hypogastrium	4 cm	US	5	EM
4	2010	34	cyclic pain	C/S	PE	hypogastrium	1 cm	US, IRM	3	E
5	2011	32	pain	C/S	tumor	hypogastrium	3 cm	US	11	EM
6	2016	42	pain	2 C/S, SG excision	SG	hypogastrium + pelvic endometriosis	2 cm	US, CT, IRM	6	E+THBA
7	2013	28	cyclic pain	2 C/S, PE excision	PE	hypogastrium	14 cm	US, IRM	7	EM+EMR
8	2015	24	pain	episiotomy	PE	perineal	4 cm	US	4	E
9	2015	28	pain	episiotomy	PE	perineal	5 cm	US	2	E+SM
10	2015	35	cyclic pain	C/S	PE	hypogastrium	5 cm	US	4	EM

C/S: cesarean section, SG: stitch granuloma, PE: parietal endometriosis, US: ultrasound, d.v.: dimensional variations, E: simple excision, EM: excision with mesh repair, M: myometrectomy, THBA: total hysterectomy with bilateral adnexectomy, SM: sphincter ani myorrhaphy

Figure 1. Small foci of abdominal wall endometriosis (arrow): after cesarean delivery scars - MRI evaluation in sagittal T1 FSE (A), axial T2 FSE (B) and T1 EG Fat Sat (C) plane

Figure 2. Involvement of rectus abdominal muscles in PE located above the Phannenstiell scar on the median line. In the right upper box: macroscopic honey-comb appearance of the resected specimen

surgical scar after cesarean section; in one case the mass appeared after hysterectomy; in two cases the mass was described at episiotomy site (*Fig. 3*); in the remaining two cases, inguinal localization was identified, in the absence of any surgical scar.

In the analyzed lot, two patients had PE associated with pelvic endometriotic lesions involving the uterus and ovaries (*Fig. 4*). The main presenting complaint for 9 of 10 patients was pain, with cyclic manifestation in four of them. All the patients presented palpable mass with tumor dimensions ranging from 1 to 14 cm (mean diameter 4.45 cm). The patient without any pain complaint had cyclic size variations of the palpable mass. One patient

Figure 3. PE at episiotomy site. In the right upper box: macroscopic honey-comb appearance of the resected specimen

accused associated dyspareunia and dysuria. Two patients had recurrence after anterior surgical interventions for PE. In two patients comorbidities were found: non-Hodgkin lymphoma (1 patient), von Willebrand disease (1 patient). All the patients were investigated by bidimensional and color Doppler ultrasound. In four cases, the diagnosis was sustained by magnetic resonance imaging (*Fig. 5*). It was

Figure 4. Endometriosis with different locations. Right ovarian cystic lesion (arrows) hyperintense on fat-suppressed T1-wi (a) and hypointense on T2-wi (b) - typical MRI findings for endometrioma; note also a very small hyperintense foci into the left ovary (arrow) and few small hyperintense spots on fat-suppressed T1 EG within the myometrium (arrows)

Figure 5. Abdominal wall endometriosis - MRI evaluation in sagittal (A), coronal (B) T2-wi and axial T1wi plane shows into the anterior abdominal wall, at the level of the surgical scar, an infiltrating heterogeneous iso-/ high-signal-intensity mass involving both muscular abdominal wall, subcutaneous fat (arrows) and uterus

performed with moderate repletion of the urinary bladder and using an intravenously administered antispasmodic agent (Buscopan). Images were acquired on a 1.5-T MRI system. Our MRI protocol included axial fat-suppressed T1 EG-weighted images (wi) in axial plane, and axial, oblique coronal, and sagittal T2-weighted sequences. Fat-suppressed T1-weighted sequences are the most sensitive for the detection of bloody foci (*Fig. 1* and *Fig. 4*), whereas high-resolution T2-weighted images are used for the evaluation of fibrotic lesions, notably those that involve the pelvic ligaments, retrocervical space, or prevesical recess. Only in one patient ultrasound, computed tomography and magnetic resonance imaging were performed all together.

Laboratory test abnormalities were found only in 2 patients that had CK-MB values elevated and respectively serum fibrinogen increased. The levels of serum tumoral markers were not checked. In half of the patients PE was diagnosed preoperatively, while four patients had preoperative diagnosis of abdominal wall tumor and one - stitch granuloma. All our patients were submitted to surgical excision of endometrioma. Three frozen section examinations were carried out during operation; two gave the diagnosis of PE, one - inflammatory tumor. In 3 cases, besides the PE resection with wide margins, other

procedures were associated to remove the involved tissues: endomyometrial resection (EMR) with hysterorrhaphy (1 patient), myorrhaphy of anal sphincter (1 patient), total hysterectomy with bilateral adnexectomy (1 patient). Four of six patients with hypogastric PE needed mesh repair of the abdominal wall. One of two patients with PE in the right groin required prosthetic material interposition. Both patients with perineal endometriosis had no need of mesh placement.

The final diagnosis of PE in all patients was made by histopathologic examination on paraffin of the surgical specimen. The margins of the specimens were confirmed to be free of disease (*Fig. 6A*). Histological examination reported the lesion involving muscle tissue (5 cases), round ligament (2 cases), subcutaneous tissue (2 cases) and one specimen associated anterior uterine wall invasion. In one patient immunohistochemistry tests were performed, showing ER-nuclear index 100% positive, PGR nuclear index 100% positive, WT1 absent in glandular epithelium, focally positive in stromal cells, CA 125 diffusely positive in glandular epithelium, Ki67 nuclear index 1-2% positive in glandular epithelial cells (*Fig. 6B-F*). The mean hospital stay was 5.2 days (4-11 days). Up to now no recurrence of PE was encountered in our patients during close postoperative follow-up.

Figure 6. (A) Endometriotic glands with stromal cells, hematoxylin & eosin stain (400X); (B) ER positive nuclear staining by immunohistochemistry (400X); (C) PR positive nuclear staining by immunohistochemistry (400X); (D) Immunohistochemistry with Ki-67 positive nuclear staining in rare cells (400X); (E) Immunohistochemistry with CA-125 positive nuclear staining in epithelium of endometriotic glands (400X); (F) Immunohistochemistry with WT1 positive nuclear staining in associated stromal cells (400X)

Discussions

The rarity of PE is attested in the literature with paucity of reported cases. However the biggest recently reported study included 227 patients treated in a gynecological department (11). General surgeons are seldom involved in treating PE, the biggest series treated in a surgical department being of only 15 cases (12). PE incidence reaches 1.08-2% after hysterotomy (13), 0.03-1% after cesarean section (14), 0.06-0.7% after episiotomy (15), values that are significantly lower than those reported in the past (16-18). This reduction in incidence is partially due to the preventive methods that gained popularity after Wasfie had suggested cleaning thoroughly and washing the incision area with saline after surgeries on uterus and uterine tubes (19). However, PE does not necessarily occur after gynecological procedures, instances of PE being observed after laparoscopic procedures, appendectomy, inguinal hernia repair, colorectal surgery (20-23) or even de novo (24). We

encountered two patients with groin masses with no previous surgical scar. PE may coexist with pelvic endometriosis in 26% of patients (3). Two of the patients in our series had associated ovarian and uterine endometriosis.

Pathogenesis of PE can be explained with several theories (25): direct inoculation during surgical interventions and subsequent stimulation by estrogens, that could be the incriminating factor in eight of our patients with previous gynecological interventions; retrograde menstruation theory (26); immune system dysfunction and autoantibody formation (similar to autoimmune diseases); coelomic metaplasia theory; lymphatic and blood dissemination of endometrial cells, that could explain the groin PE in two of our patients; stem cell theory; embryonic rest theory that explains the endometriosis presence in men (27,28). Alcohol consumption and heavy menstrual flow may act as predisposing factors to disease occurrence. Instead, high parity is considered to be a protective factor against PE (29). Clinically, PE usually respects the triad

described by Esquivel: palpable mass, catamenial pain and history of cesarean section (30). Cyclicity of pain is not found in all cases. Other symptoms such as increasing size of lump, bleeding, discharge, skin discoloration, dysmenorrhea, infertility and dyspareunia are mentioned (4). In our study, catamenial pain was rated as only 40%, which could explain the rate of 50% misdiagnoses before operation. The accuracy of preoperative diagnosis is reported between 20% and 50% (31). One patient had the mass and cyclical pain located away from the Pfannenstiel incision, supposedly because of the vertical incision of the peritoneum. Time interval elapsed from last operation and presentation could range from 6 months to 10 years (9). Our study confirmed the literature results, with a preoperative diagnosis of PE in half of the cases. This denotes that thorough supplementary investigations should be performed. Abdominal sonography is the most cost-efficient investigation. It may reveal hypoechoic inhomogenous echo-texture with scattered hyperechoic echoes and irregular margins, often spiculated, surrounded by a hyperechoic ring of variable width and continuity. On color Doppler examination, single pedicles may be identified at periphery of the mass (32). If the lesion is sizable or extending beyond abdominal wall, then the patients should undergo MRI, in order to better characterize the mass anatomy and its surrounding structures (33). The major role of MRI is to depict the extent of the disease preoperatively. MR characteristics of PE may be nonspecific, showing a solid enhancing mass in the abdominal wall (34). In cases in which MRI shows a hyperintense heterogenous lesion associated with anterior abdominal or pelvic wall surgical scarring on both T1- and T2-weighted images, subacute hemorrhage within endometrioma may be the reason (35). Imagistic results may be helpful in surgical planning to estimate the parietal defect and need of mesh parietal repair (8). In our study, MRI was useful for evaluating the depth of infiltration in 40% of patients. Although fine-needle aspiration cytodiagnosis was used by other authors and gave a precise diagnosis, its application

remains controversial because of risk of endometrioid tissue dissemination (36). The serum level of CA-125 was reported in the literature to be normal or slightly increased (20). Because PE is easily confound with surgical lesions like umbilical and inguinal hernias, abscess, stitch granuloma, hematoma, tumors, because imagistic methods are non-specific, and, moreover, some patients have no antecedents of operations, histopathological analysis of the surgical specimen is mandatory (24). The histopathological diagnosis was performed in all our patients and was based on recognition of at least 2 from 3 criteria: endo-metrial stroma, endometrial glands and hemosiderin pigment (37). Medical treatment with combined oral contraceptives, progestogens (dienogest), anti-estrogens (danazol), gonadotropin-releasing hormone agonists (triptoreline, gosereline) or aromatase inhibitors (letrozol) provides temporary alleviation of symptoms with their recurrence after drug cessation (11) and could be recommended to prevent recurrences after PE resection. Surgical excision of PE is unanimously considered to be the only curative treatment. However, less invasive, in situ destructive methods like cryoablation (38), high intensity focus ultrasound (39) or ethanol injection (40) have been reported, but more time should pass to decide upon their safety and efficacy. It is preferred to conduct the operation under general anesthesia. Wide surgical excision with at least 1 cm margin resection is advisable. Sometimes preoperative placement of a hook wire under ultrasound guidance is useful in tracking small, impalpable lesions (41). If the lesion is large and extends to deeper tissues such as muscle, aponeurosis or peritoneum and even invades other organs, visceral resections and reconstructive techniques may be required (42). In our series, 50% of the patients had invasion of muscular tissue, 20% - round ligament and 10% - uterus. Mesh repair of the anterior abdominal wall was necessary in 50% of patients. Another useful surgical technique for anterior wall reconstruction is rotation of aponeurotic muscle flap (43). The involvement of the anal sphincter is a rare occurrence in perineal endometriosis. The clinical bimanual

simultaneous examination of vagina and rectum is important to appreciate the local extension of the mass. In case of sphincter involvement, anal manometry and endoanal ultrasound are recommended. The surgical procedure implies removal of the lesion with part of the external anal sphincter followed by its reconstruction (e.g. “overlapping” fashion, “apposition” technique) (44). For one of our patients with endometriosis at the level of episiotomy scar, a careful dissection of the nodule was done in order to preserve as much as possible from the external anal sphincter while removing all the lesion with free margin of at least 0,5 cm; the excision was followed by myorraphy of levator ani to secure the perineal body. Recurrence after surgical excision is rare in PE, if properly diagnosed and treated, as well as its malignant transformation (45).

Conclusions

Even if rarely encountered by the general surgeon, PE must be thought in all female patients with/without previous surgical or obstetrical interventions, that complain of catamenial pain at the level of abdominal wall or perineum. The diagnosis should be sustained by imagistic studies and confirmed by histopathology. The only curative treatment remains large excision of PE with disease-free margins under general anesthesia. Mesh repair of abdominal wall and partial resection with/without reconstruction of anal sphincter are sometimes required and fall into the competence of general surgery. Insufficient awareness of general surgeons about PE may lead to misdiagnosis and improper treatment, that may result in prolonged suffering of the patient.

Conflict of Interest

All author declare that they have no conflict of interest.

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