

RUPTURED OVARIAN ENDOMETRIOTIC CYST IN A 28-YEAR-OLD WOMAN - CASE REPORT

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ABSTRACT:

ENDOMETRIOSIS IS A COMMON BENIGN DISEASE, WITH DIFFICULT QUANTIFICATION OF INCIDENCE GIVEN BY THE DELICATE SONOGRAPHIC DIAGNOSIS, WITH LOW SENSITIVITY FOR SMALLER IMPLANTS. THE MAIN METHOD OF DIAGNOSIS IS LAPAROSCOPIC SURGERY WITH BIOPSY AND HISTOPATHOLOGY POSITIVE RESULT. USING THIS STANDARD 1.6/1000 WOMEN BETWEEN 15 AND 49 SUFFER FROM ENDOMETRIOSIS, BUT THE UNDIAGNOSED CASES ARE PROBABLY MUCH MORE. PRIOR, IT WAS BELIEVED THAT WHITE WOMEN REPRESENTED THE MOST AFFECTED ETHNICITY, BUT RECENTLY VARIABLE RESULTS WERE OBSERVED. WE REPORT A CASE OF A 28-YEAR-OLD WOMAN WITH A HISTORY OF ENDOMETRIOTIC CYST WHO PRESENTED AT THE EMERGENCY ROOM WITH ACUTE ABDOMEN AFTER SEXUAL INTERCOURSE. MAKING A REVIEW OF THE LITERATURE, WE FOUND FEW CASES OF RUPTURED LARGE ENDOMETRIOTIC CYSTS. THE POSTOPERATIVE EVOLUTION OF THE PATIENT WAS FAVORABLE, WITH GOOD RESPONSE OF TREATMENT AT LATER FOLLOW-UP.

KEY WORDS: ENDOMETRIOTIC OVARIAN CYST, ENDOMETRIOSIS, OVARIAN CYST

INTRODUCTION

Endometriosis represents the presence of endometrial glands and stroma in other locations than the uterine cavity¹³. Such locations with abnormal implants are pelvic peritoneum, ovaries, uterosacral ligaments as the most common ones¹⁴.

Even though endometriosis is frequently a nonmalignant process, such abnormal implantations of endometrial tissue can cause chronic pain, dysmenorrhea, dyspareunia and infertility¹⁵, many of them also present in other conditions. This symptoms can even reach paralyzing pain, affecting the day to day life of the patient¹⁶. It is an estrogen-dependent, inflammatory illness, which affects women during their premenarcheal, reproductive, and postmenopausal hormonal stages¹⁷. Not much is known about prevalence and risk factors when it comes to endometriosis, accurate assessment of epidemiological findings of these disease is held back by the incapacity to detect it in general population¹⁸. There are some theories when it comes to pathogenesis such as retrograde menstruation, coelomic metaplasia and Müllerian remnants,

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¹⁸ Eskenazi, B; Warner, ML; *Epidemiology of endometriosis*. Obstet Gynecol Clin North Am 1997; 24:235.

neither explaining all types of endometriosis¹⁹. Risk factors may include nulliparity, low BMI²⁰, late menopause, shorter menstrual cycle (< 27days), heavy menstrual bleeding, obstruction of menstrual outflow²¹, early menarche²², sexual abuse as child or teenager²³, high intake of trans saturated fat²⁴.

Clinical examinations in woman with endometriosis is variable, sometimes the clinician can observe vaginal tenderness, fornix nodules, pelvic mass which may place the uterus laterally²⁵.

There are no specific laboratory findings, but as a routine all woman suspected of endometrioma are required CA125 marker, which can be elevated²⁶.

For preoperative diagnoses sonography magnetic resonance imaging can be used to detect ovarian cysts, rectovaginal nodules and bladder nodules²⁷, some abdomen wall lesion can be seen as hypoechogenic vascular lesions with irregular outline²⁸.

Considering symptoms, response to medication, clinical and paraclinical findings, sometimes surgery is required, laparoscopic surgery is usually the preferred method but emergency situations may indicate laparotomy²⁹. Staging: I - minimal disease is characterized by isolated implants and no significant adhesions, II - superficial implants that are less than 5 cm in aggregate and are scattered on the peritoneum and ovaries, no significant adhesions are present, III - both superficial and deeply invasive, peritubal and periovarian adhesions may be evident, IV - multiple superficial and deep implants, including large ovarian endometriomas, filmy and dense adhesions are usually present³⁰.

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²¹ Sinaii, N; Plumb, K; Cotton, L; et al. *Differences in characteristics among 1,000 women with endometriosis based on extent of disease*. Fertil Steril 2008; 89:538; Treloar, SA; Bell, TA; Nagle, CM; et al. *Early menstrual characteristics associated with subsequent diagnosis of endometriosis*. Am J Obstet Gynecol 2010; 202:534.e1; Giudice, LC; *Clinical practice. Endometriosis*. N Engl J Med 2010; 362:2389.

²² Nnoaham, KE; Webster, P; Kumbang, J; et al. *Is early age at menarche a risk factor for endometriosis? A systematic review and meta-analysis of case-control studies*. Fertil Steril 2012; 98:702.

²³ Harris, HR; Wieser, F; Vitonis, AF; et al. *Early life abuse and risk of endometriosis*. Hum Reprod 2018; 33:1657.

²⁴ Missmer, SA; Chavarro, JE; Malspeis, S; et al. *A prospective study of dietary fat consumption and endometriosis risk*. Hum Reprod 2010; 25:1528.

²⁵ Hickey, M; Ballard, K; Farquhar, C; *Endometriosis*. BMJ 2014; 348:g1752.

²⁶ Mol, BW; Bayram, N; Lijmer, JG; et al. *The performance of CA-125 measurement in the detection of endometriosis: a meta-analysis*. Fertil Steril 1998; 70:1101.

²⁷ Guerriero, S; Saba, L; Pascual, MA; et al. *Transvaginal ultrasound vs magnetic resonance imaging for diagnosing deep infiltrating endometriosis: systematic review and meta-analysis*. Ultrasound Obstet Gynecol 2018; 51:586.

²⁸ Hensen, JH; Van Breda Vriesman, AC; Puylaert, JB; *Abdominal wall endometriosis: clinical presentation and imaging features with emphasis on sonography*. AJR Am J Roentgenol 2006; 186:616.

²⁹ Pardanani, S; Barbieri, RL; *The gold standard for the surgical diagnosis of endometriosis: Visual findings or biopsy results?* J Gynecological Techniques 1998; 4:121; Almeida Filho, DP; Oliveira, LJ; Amaral, VF; *Accuracy of laparoscopy for assessing patients with endometriosis*. Sao Paulo Med J 2008; 126:305.

³⁰ *Revised American Society for Reproductive Medicine classification of endometriosis: 1996*. Fertil Steril 1997; 67:817.

Endometriotic foci most often appear as punctate red, brown or white areas³¹. The age and functional state determines their colour as following: the earliest lesions present as yellow-red surface stains, while the red ones are also early forms of the disease, only actively growing; these are followed by black lesions, most commonly seen by pathologist in operation specimens, in which the bleeding has resolved; some lesions can be brown to slightly yellow, reflecting hemosiderin deposits, while the oldest of them are white, with fibrosis and scarring³². At some sites, mostly in the ovary, lesions can become cystic and reach impressive sizes, with diameters up to 15cm³³. Endometriotic cysts (endometriomas) have fibrotic walls with a smooth or ragged brown to yellow lining and semifluid or thickened brown, chocolate-coloured contents³⁴.

Microscopically, endometriosis consists of endometrial glands and stroma, with the diagnosis being often possible with only component identified³⁵; however, they should both be definitively identified for avoidance of diagnosis errors. The glandular epithelium is one cell thick consisting of columnar cells, with cigar-shaped, vertically oriented nuclei and eosinophilic cytoplasm; cilia can sometimes be identified. Serous (tubal), clear cell, mucinous and even squamous metaplasia can occur³⁶. Usually, endometrial-type glands are associated with fibrosis, hemosiderin deposits and histiocytes³⁷. The stroma is composed of small, elongated cells with scant cytoplasm and a delicate reticulin network³⁸. Decidualization of the stroma and gestational changes can occur.

Endometriomas usually feature an extremely attenuated epithelial lining who can be totally replaced by granulation tissue, fibrous tissue and pseudoxanthoma cells³⁹. These findings along with hemosiderin deposits and islands of residual cuboidal glandular epithelium or endometrial stroma can lead to a diagnosis of endometriotic cyst.

The most important entities to be considered in the differential diagnosis of endometriosis are solitary follicle or corpus luteum cysts, tubo-ovarian abscesses, surface epithelial cysts/cystadenomas, unilocular cystic granulosa cell tumours and secondary neoplasm (especially

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³² Mutter, George L; Pratt, Jaime; *Pathology of the Female Reproductive Tract*, 3rd edition, 2014, Churchill Livingstone.

³³ Clement, Phillip; Stall, Jennifer; Young, Robert; *Atlas of Gynecologic Surgical Pathology*, 4th Edition, 2019, Elsevier; Mutter, George L; Pratt, Jaime; *Pathology of the Female Reproductive Tract*, 3rd edition, 2014, Churchill Livingstone.

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³⁸ Mutter, George L; Pratt, Jaime; *Pathology of the Female Reproductive Tract*, 3rd edition, 2014, Churchill Livingstone; Evangelinakis, N; Grammatikakis, I; Salamalekis, G; et al. *Prevalence of acute hemoperitoneum in patients with endometriotic ovarian cysts: a 7-year retrospective study*. Clin Exp Obstet Gynecol. 2009;36:254–255.

³⁹ Clement, Phillip; Stall, Jennifer; Young, Robert; *Atlas of Gynecologic Surgical Pathology*, 4th Edition, 2019, Elsevier.

clear cell or endometrioid carcinoma). Immunohistochemically, the epithelium of the endometriotic glands is positive for ER and PR, while the stroma shows typical positivity for ER, PR and CD10.

Malignancy can arise in up to 1% of the cases, the most common being clear cell and endometrioid carcinomas⁴⁰.

Generally the prognosis of patients with endometriosis is good. It is important to understand the high possibility of recurrence and the long term medical treatment, as it is considered a chronic disease.

Treatment is a complex problem in this pathology, depending on the age, symptoms and fertility preservation. It may include medical treatment of the pain with nonsteroidal antiinflammatory drugs, combination hormonal contraceptives, progestins, GnRH agonists, GnRH antagonists, aromatase inhibitors, selective progesterone – receptor modulators, androgens or surgery⁴¹.

Patients are advised to have a long-term follow-up.

The aim of this case report is to show the diagnostic stages and treatment of a rare event, which is encountered in less than 3% of women of childbearing age who are known to have endometriomas⁴².

Our case was appealing for 2 reasons: the size of the endometrioma with thick wall and the fact that it ruptured.

CASE PRESENTATION

A 28-year-old woman, C.A.M., was admitted in the Department of Obstetrics and Gynaecology “St. Pantelimon” Emergency Clinical Hospital, Bucharest, Romania, on June 2017, for acute abdominal pain and muscular defense after intercourse in order to determine therapeutic specialist conduct. From the patient's personal history, we take note of 0 pregnancies and bilateral ovarian endometriomas.

At admission in our department, the patient was cooperative, blood pressure was 130/75 mmHg, pulse 110 beats/minute. The gynaecological examination with the speculum showed tenderness, no macroscopic lesions on the cervix and no bleeding coming from the uterine cavity. On the bimanual examination we could identify the uterus, in intermediary position, highly painful with mild palpation, with difficulties in assessing the size, right adnexa increased in volume, highly painful at palpation, with impossible to determine shape and consistency because of muscle defense, left adnexa painful at mild palpation, increased in volume, vaginal fornix bulges. Laboratory exams were completed and were in normal range. Transvaginal ultrasound was performed and revealed uterus in intermediary position, surrounded by hypoechoic fluid, on the right of the uterus and continuing to the Douglas an ovoidal formation can be seen, with irregular shape, thick wall and non-homogeneous hypoechoic content; on the left ovary a cystic formation

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with a 3 cm diameter and non-homogeneous hypochoic content could be observed (Figure 1., Figure 2., Figure 3., Figure 4.).



Figure 1) Transvaginal sonography.



Figure 2) Transvaginal sonography.



Figure 3) Transvaginal sonography.

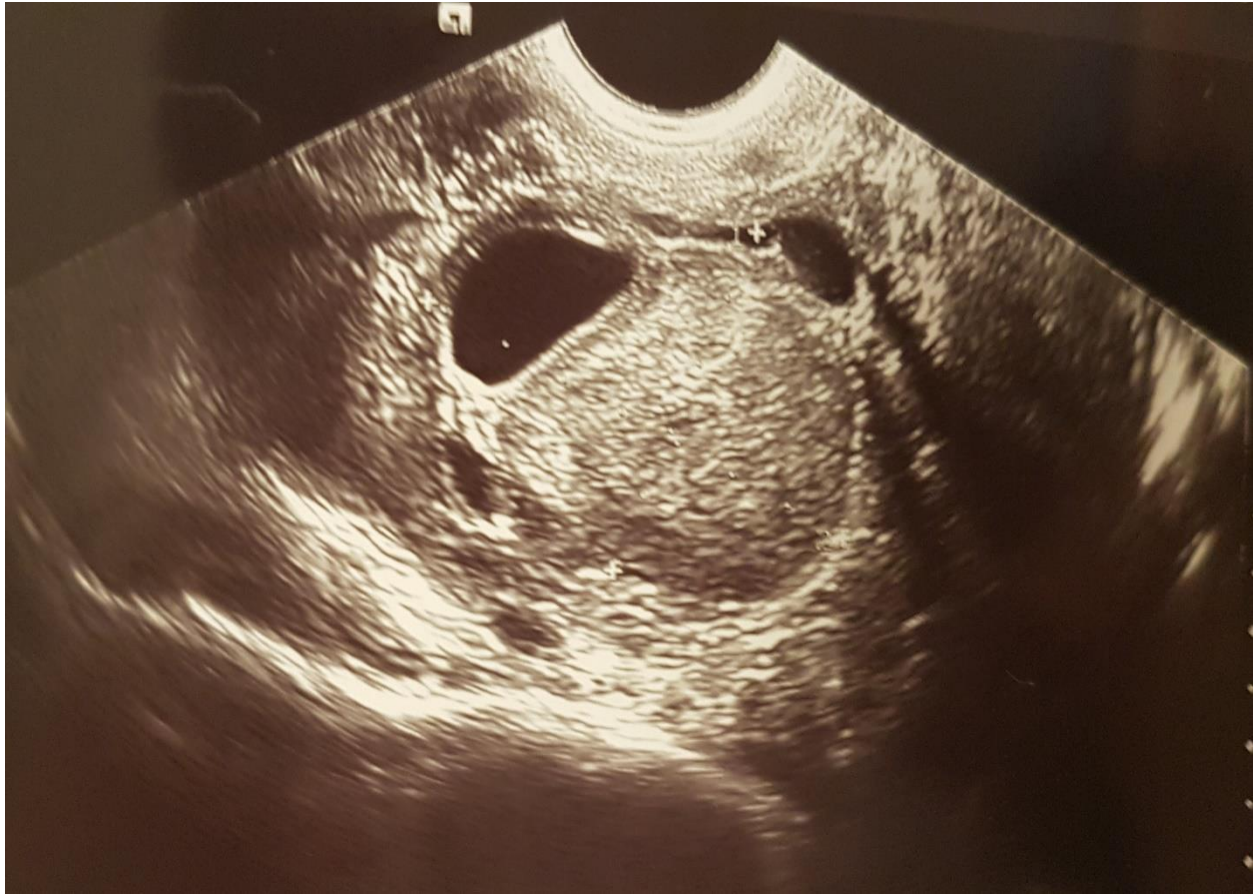


Figure 4) Transvaginal sonography.

The informed consent was obtained and exploratory surgery through laparotomy was performed under general anaesthesia, followed by aspiration of 400 ml of blood and chocolate-like fluid. Upon inspection of the peritoneal cavity on the right side of the uterus one could observe a large ruptured ovarian endometrioma, approx. 15 cm in diameter, adherent to the peritoneum, uterus and rectum; on the left side there was an intact 3 cm ovarian endometrioma. The surgical team decide to perform right cystectomy and left cystectomy and send the specimens for histopathological evaluation.

The postoperative evolution was favourable, without any complications. On day 5, the patient was released from the hospital with good general condition and afebrile. Progestins were given for the next 6 months after surgery.

The histopathological result showed a flattened columnar epithelial lining and typical endometriotic stroma with hemosiderin deposits and foci of granulation tissue.

At the 6 months follow-up of the patient - no complications occurred and the serum investigations were in normal limit. Progestins were replaced with combination birth control until the patient wishes for a pregnancy.

DISCUSSION

Clinical diagnosis is difficult if no sonographic signs are discovered⁴³. Most of the times the patient presents symptoms which are described in other conditions as well and differential diagnoses is difficult to make. In our case the patient was had a known history of ovarian endometriomas and presented with acute abdomen and a positive sonographic diagnose which led to emergency laparotomy.

It is preferred to exclude the endometrioma without rupturing it, to prevent spreading the content in the peritoneal cavity as it is believed this increases the likelihood of a relapse. In our case it was not possible and lavage with sodium chloride was done multiple times.

Periodical reevaluation in such cases is mandatory as this is a chronic disease with a positive outcome if managed correctly.

Conduction of patients with infertility and wish for a pregnancy is difficult due to the higher possibility of having a more severe extension of the disease. Managing such cases and obtaining a pregnancy has proven to be a difficult task. Severe cases (stage IV) are able to obtain pregnancies by in vitro fertilisation, with poorer implantation rates than stage III⁴⁴.

CONCLUSIONS

Ruptured ovarian endometriomas are rare cases which must be treated carefully by experienced gynaecologists. Surgery is required and the type of the surgery is selected by the doctor taking into consideration the experience and the size of the ruptured endometrioma. CA125 may be elevated and the extracted specimen must be sent to the histopathologic department to exclude other possibilities and correctly conduct treatment afterwards.

Fertility can be preserved, but caution must be implied. The majority of the patients have a good outcome.

⁴³ Stanimir, M; Chiutu, LC; Wese, S; Milulescu, A; Nemes, RN; Bratu, O. *Mullerianosis of the urinary bladder: a rare case report and review of the literature*. Rom J Morphol Embryol. 2016; 57(2 Suppl): 849-852; Socea, LI; Visan, DC; Barbuceanu, SF; Apostol, TV; Bratu, OG; Socea, B. *The antioxidant activity of some acylhydrazones with dibenzo[a,d][7]annulene moiety*. Rev Chim (Bucharest), 2018, 69(4): 795-797; Bodean, O; Bratu, O; Bohiltea, R; Munteanu, O; Marcu, D; Spinu, DA; Vacarioiu, IA; Socea, B; Diaconu, CC; Fometescu Gradinaru, D; Cirstoiu, M. *The efficacy of synthetic oral progestin pills in patients with severe endometriosis*. Rev Chim (Bucharest), 2018, 69(6): 1411-1415; Bratu, OG; Marcu, RD; Socea, B; Neagu, TP; Diaconu, CC; Scarneci, I; Turcu, FL; Radavoi, GD; Bratila, E; Berceanu, C; Spinu, AD. *Immunohistochemistry particularities of retroperitoneal tumors*. Rev Chim (Bucharest), 2018, 69(7): 1813-1816; Dimitriu, MCT; Ionescu, CA; Gheorghiu, DC; Socea, LI; Bratu, OG; Constantin, VD; Ples, L; Neacsu, A; Bobic, S; Socea, B. *Mepivacaine hydrochloride -an efficient local anesthetic solution for the electroresection of the benign and preneoplastic lesions of the cervix and uterus*, Rev Chim (Bucharest), 2018, 69(9): 2391-2395; Bodean, O; Bratu, O; Munteanu, O; Marcu, D; Spinu, DA; Socea, B; Diaconu, C; Cirstoiu, M. *Iatrogenic injury of the low urinary tract in women undergoing pelvic surgical interventions*. Archives of the Balkan Medical Union, 2018, 53(2): 281-284.

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ACKNOWLEDGEMENTS

All authors equally contributed in the research and drafting of this paper.

All authors report no potential conflict of interest.

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