

1 HEMOPERITONEUM FOLLOWING SEXUAL INTERCOURSE  
2 IN A WOMAN WITH DEEP INFILTRATING ENDOMETRIOSIS  
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## 24 ABSTRACT

25 Endometriosis is an estrogen-dependent chronic inflammatory disease, defined by presence of  
26 endometrial glands and stroma at ectopic sites. A rare and life-threatening complication associated  
27 with endometriosis is represented by spontaneous hemoperitoneum due to the rupture of utero-  
28 ovarian vessels. Most cases of spontaneous hemoperitoneum previously described involved  
29 pregnant women affected by endometriosis; here we present a case of acute and massive  
30 hemoperitoneum in a nulliparous woman with deep infiltrating endometriosis. When acute  
31 abdominal pain with hemoperitoneum occurs in non-gravid reproductive age women, with no  
32 positive findings for liver or spleen lesions, a possible spontaneous rupture of utero-ovarian vessels  
33 related to the presence of deep infiltrating endometriosis should be included among the possible  
34 causes of the condition.

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## 36 ESTABLISHED FACTS

37 - A rare and life-threatening complication associated with endometriosis is represented by  
38 spontaneous hemoperitoneum.

39 - Chronic inflammation associated with deep infiltrating endometriosis may render pelvic vessels  
40 more friable and prone to spontaneous rupture following a stretching insult.

## 41 NOVEL INSIGHTS

42 - When acute abdominal pain with hemoperitoneum occurs in non-gravid reproductive age women,  
43 with no positive findings for liver or spleen lesions, a possible spontaneous rupture of utero-ovarian  
44 vessels related to the presence of deep infiltrating endometriosis should be included among the  
45 possible causes of the condition.

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## 47 INTRODUCTION

48 Endometriosis is an estrogen-dependent chronic inflammatory disease, defined by presence of  
49 endometrial glands and stroma at ectopic sites; this condition affects women in their reproductive  
50 period and is associated with different clinical symptoms, such as dysmenorrhea, dyspareunia and  
51 chronic pelvic pain [1].

52 A rare and life-threatening complication associated with endometriosis is represented by  
53 spontaneous hemoperitoneum; most of the cases previously reported in the literature described  
54 events of spontaneous hemoperitoneum in pregnant women with endometriosis [2-4]. Spontaneous  
55 hemoperitoneum typically presents with signs of acute intraperitoneal bleeding and occasionally  
56 abdominal compartment syndrome in severe cases [4].

57 Here we present a case of acute and massive hemoperitoneum in a nulliparous woman affected by  
58 deep infiltrating endometriosis.

## 59 CASE REPORT

60 A 45-year-old nulliparous woman was admitted to the emergency department with acute diffuse  
61 lower abdomen pain and asthenia. Her medical history was unremarkable, and she denied previous  
62 surgery. She was assuming oral contraceptives and reported a sexual intercourse the night before  
63 the admittance.

64 An initial physical examination revealed a distended abdomen, tenderness to abdominal palpation  
65 without acute peritoneal signs (negative Blumberg sign), and no vaginal bleeding; the clinical vital  
66 signs were stable (blood pressure 107/55 mmHg, pulse rate 120 beats/min, oxygen saturation 99%,  
67 normal body temperature). A urine pregnancy test was negative.

68 The hemogasanalysis showed a severe decrease of hemoglobin level (Hb = 3.8 g/dl). The  
69 subsequent laboratory tests confirmed the severe anemia with a hemoglobin level of 3.5 g/dl and a  
70 hematocrit of 10.9%. Due to the results of the exams the patient was immediately transfused with 3  
71 units of red blood cells.

72 An emergency computerized tomography (CT) demonstrated the presence of a significant amount  
73 of free intraperitoneal fluid, suggesting an internal bleeding, moreover the CT scan reported an  
74 inhomogeneous aspect of the uterine fundus, without clear identification of a source of active  
75 bleeding.

76 Therefore, the patient was transferred to our obstetric-gynecological emergency department in order  
77 to perform a gynecological examination. The abdominal and transvaginal ultrasound revealed  
78 evidence of free fluid in the abdomen and tenderness of the Douglas pouch at the mobilization of  
79 the vaginal probe, the ovaries were poorly evaluable for the presence of blood clots, moreover free  
80 fluid was observable in both the left and right hypochondrium. In the mean time, the general  
81 conditions of the patient were worsening, with the appearance of unstable clinical signs, and  
82 positive peritoneal signs at the the physical examination. The laboratory tests following the blood  
83 transfusions still showed low hemoglobin (7.6 g/dl) and hematocrit (22.0%) levels.

84 Considering the worsening conditions of the patient an exploratory laparoscopy was performed.  
85 Laparoscopy revealed about 2800 mL of fresh liquid and clotted blood; after aspiration of the  
86 blood, the uterus appeared tenaciously adherent to the anterior rectal wall with obliteration of the  
87 pouch of Douglas. In addition, the ovaries were bilaterally densely adherent to the broad ligaments,  
88 the rectum and the uterus. After partial adhesiolysis, the surgeon identified a fissure of the left  
89 uterine artery as the source of the bleeding. No sign of vessels' decidualization could be identified.  
90 Hemostasis was obtained applying bipolar coagulation to the bleeding artery; moreover, in order to  
91 achieve better hemostasis, a hemostatic matrix was used. At the end of the procedure, biopsies of  
92 the left ovary and of the peritoneal lesions were obtained.

93 The above picture depicts a severe deep infiltrating endometriosis form, and the surgical diagnosis  
94 was eventually confirmed by the results of the histopathologic examination of the specimens.

95 The patient received 7 units of red blood cells and remained stable after surgery, with a hemoglobin  
96 value at discharge of 11.0 g/dL and a hematocrit of 32.4 %. In postoperative day 4 a gynecological  
97 evaluation was performed; the transvaginal ultrasound revealed a finely inhomogeneous

98 echogenicity of the uterus suggestive for a condition of diffuse adenomyosis and detected the  
99 presence of adhesions between the serous of the rectum and the posterior uterine wall in  
100 correspondence of the “torus uterinus”, attributable to a consequence of pelvic endometriosis.  
101 The patient was discharged on postoperative day 5 in stable conditions. The follow-up control at 40  
102 days from surgery was normal.

### 103 DISCUSSION

104 As recently reported in a systematic review by Viganò *et al.* [3] endometriosis represent a major  
105 risk factor for spontaneous hemoperitoneum in pregnancy, with an overall prevalence of this  
106 complication of 0.4%. However, only few cases of non-gravid women with endometriosis and  
107 spontaneous hemoperitoneum have been previously described [6-14] (Table 1).

108 In our case the patient reported a vigorous sexual intercourse immediately before insurgence of the  
109 abdominal pain. Chronic inflammation associated with deep infiltrating endometriosis may render  
110 pelvic vessels more friable and prone to spontaneous rupture following a stretching insult, as it is  
111 the case during pregnancy the uterus enlarges or, in non pregnant conditions, after a sexual  
112 intercourse. More in general, activities like coughing, defecation, muscular activity or coitus  
113 increase venous pressure in the utero-ovarian circulation, thus increasing also the risk of venous  
114 vessels rupture [4].

115 Given the rarity of this complication a prompt diagnosis appears of primary importance, in fact in  
116 the majority of the cases of massive hemoperitoneum the patient reaches the hospital with severe  
117 abdominal pain associated with hypovolemic shock and acute anemia. In addition, hemothorax  
118 represents a possible associated atypical sign [3]. Imaging techniques for the diagnosis of SH  
119 comprehend ultrasonography and, secondly, computerized tomography.

120 If possible, a pre-operative diagnosis of the source of the bleeding should be performed; the  
121 spontaneous rupture of a utero-ovarian vessel enters in differential diagnosis with other causes of  
122 spontaneous hemoperitoneum, like rupture of an ectopic pregnancy or a corpus luteum cyst, rupture  
123 of an adenomyotic uterus or leiomyomas, and of lesion of the liver or spleen.

124 In hemodynamically stable patients angiography and embolization may be considered [5].  
125 However, in case of hemorrhagic shock immediate surgical exploration is mandatory.  
126 In conclusion, when acute abdominal pain with hemoperitoneum occurs in non-gravid reproductive  
127 age women, with no positive findings for liver or spleen lesions, a possible spontaneous rupture of  
128 utero-ovarian vessels related to the presence of previously unrecognized deep infiltrating  
129 endometriosis should be included among the possible causes of the condition.  
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Table 1. Characteristics of non-pregnant women with endometriosis-associated acute hemoperitoneum. Literature data 1970-2016.

Source	Year	No. of patients	Previous diagnosis of endometriosis	Treatment	Surgical findings
Ranney B. (6)	1970	4	No	Laparotomy	Case 1. Bleeding endometriotic peritoneal deposits Case 2. Ruptured right endometrioma of 20-25 cm Case 3. Ruptured left endometrioma Case 4. Bleeding endometriotic peritoneal deposits
Carmichael <i>et al.</i> (7)	1972	1	No	Laparotomy	Active bleeding from endometriosis deposits of the posterior surface of the left broad ligament
Kumar S. (8)	1996	1	No	Laparotomy	Active bleeding from endometriotic deposits in the pouch of Douglas and along both uterosacral ligaments
Harmanli <i>et al.</i> (9)	1998	1	No	Laparotomy	Tubal endometriosis implants and a large, ruptured, hemorrhagic endometriotic cyst
Janicki <i>et al.</i> (10)	2002	1	Yes (stage III endometriosis diagnosed at prior laparoscopy)	Laparotomy	Left endometrioma (5x6 cm) Erosion of the left uterine artery

Table 1. Continued

Source	Year	No. of patients	Previous diagnosis of endometriosis	Treatment	Surgical findings
Fiadjoe <i>et al.</i> (11)	2008	1	Not specified	Laparotomy	Ruptured right endometrioma eroding into the broad ligament, pelvic side wall and uterine artery
Evangelinakis <i>et al.</i> (12)	2009	16	Not specified	Laparoscopy	Ruptured left ( $n = 11$ ) and right endometrioma ( $n = 5$ )
Mutihir <i>et al.</i> (13)	2010	1	Yes (umbilical endometriosis)	Laparotomy	Active bleeding from endometriotic deposits in the pouch of Douglas and of the sigmoid colon
Togami <i>et al.</i> (14)	2015	1	Yes (site and prior procedures not specified)	Laparoscopy	Active bleeding from endometriotic peritoneal deposits
Present report	2016	1	No	Laparoscopy	Fissured left uterine artery due to the presence of deep infiltrating endometriosis