

Case Report



Spontaneous Rupture of the Utero-Ovarian Vessels in a Non-Pregnant Woman 15 Years Following Endometriosis Eradication

Alessandro Libretti ^{1,2,*}, Anthony Nicosia ^{1,2}, Valentino Remorgida ^{1,2} and Livio Leo ³

- ¹ Department of Gynaecology and Obstetrics, University Hospital Maggiore della Carità, 28100 Novara, Italy; nicosiaanthony@hotmail.it (A.N.); valentino.remorgida@uniupo.it (V.R.)
- ² School of Gynaecology and Obstetrics, University of Eastern Piedmont, 28100 Novara, Italy
- ³ Department of Gynecology and Obstetrics, Hopital Beauregard, AUSL Valleè d'Aoste, 11100 Aosta, Italy; lleo@ausl.vda.it
- * Correspondence: libretti.a@gmail.com; Tel.: +39-347-5916561

Abstract: Spontaneous rupture of the utero-ovarian vessels is an exceptionally rare but potentially life-threatening condition, especially in the case of non-pregnant women with a history of complex gynecological conditions. We report the case of a 45-year-old woman presenting with severe abdominal pain and hemoperitoneum, 15 years after surgical eradication of stage IV endometriosis. Diagnostic imaging revealed significant free fluid and vascular disruption near the uterus. Emergency laparotomy confirmed blood in the peritoneal cavity and identified a rupture of the left paracervical vessels. This case underscores the critical role of timely surgical intervention and the challenges of diagnosing spontaneous vascular rupture in the context of chronic conditions such as endometriosis and fibromyal-gia. A review of the literature revealed very limited cases with similar presentations, emphasizing the rarity of such vascular events, although without active endometriotic lesions. This report highlights the importance of considering spontaneous vascular rupture in differential diagnoses for acute abdominal pain with hemoperitoneum. Advanced imaging and multidisciplinary management are pivotal in ensuring favorable outcomes.

Keywords: spontaneous hemoperitoneum; endometriosis; case report; laparotomy; gynecology

1. Introduction

Endometriosis is a chronic pathological condition characterized by the spread and growth of endometrial tissue (glands and stroma) in ectopic locations. It is defined as a progressive and chronic condition because these areas respond cyclically to hormonal stimuli in a similar manner to normal endometrium [1]. It is estimated to affect 6–10% of women of reproductive age worldwide [2]. Although endometriosis is a non-cancerous condition, it exhibits several characteristics commonly associated with malignant diseases, including neoangiogenesis, cellular proliferation, stromal and cellular invasion, and the capacity for dissemination. These features underscore its complex pathophysiology. Moreover, there is a notable increased risk for ovarian cancer in patients with endometriosis [1,2].

Although pelvic pain is common in women with endometriosis, it is not sufficient on its own for a diagnosis, as it may be associated with other gynecological and nongynecological conditions [3]. Some of the symptoms that accompany chronic pelvic pain include dyschezia, dyspareunia, and dysuria, which are often associated with deep endometriosis, along with lower back pain and dysmenorrhea [4]. In 30% of cases, en-



Received: 29 December 2024 Revised: 8 January 2025 Accepted: 20 January 2025 Published: 22 January 2025

Citation: Libretti, A.; Nicosia, A.; Remorgida, V.; Leo, L. Spontaneous Rupture of the Utero-Ovarian Vessels in a Non-Pregnant Woman 15 Years Following Endometriosis Eradication. *Women* 2025, *5*, 1. https://doi.org/ 10.3390/women5010001

Copyright: © 2025 by the authors. Licensee MDPI, Basel, Switzerland. This article is an open access article distributed under the terms and conditions of the Creative Commons Attribution (CC BY) license (https://creativecommons.org/ licenses/by/4.0/). dometriosis is asymptomatic and is incidentally diagnosed during a laparoscopic or open surgery performed for unexplained infertility [3].

Women with endometriosis have an increased risk of chronic pain conditions such as fibromyalgia [5–7] and migraines [8], as well as autoimmune diseases like rheumatoid arthritis and psoriatic arthritis [9,10].

Medical treatment for endometriosis aims to suppress ovarian activity and induce atrophy of endometriotic lesions. It is recommended in cases of suspected endometriosis, particularly for younger patients or those with mild symptoms. It is also appropriate when a patient opts against invasive diagnostic procedures, as surgery in these cases would typically serve both diagnostic and therapeutic purposes [11,12].

In recent years, advanced operative laparoscopic techniques have been utilized for the excision of endometriotic implants and the lysis of adhesions to relieve pain and restore normal pelvic anatomy, thereby addressing infertility [13].

Vascular complications in endometriosis are rare but can include hemoperitoneum, arterial or venous bleeding, and vascular involvement in endometriotic lesions. These complications arise due to the infiltration of endometrial-like tissue into vascular structures, leading to rupture, thrombosis, or vascular obstruction [14].

One of the key challenges in managing endometriosis is the recurrence of the disease. On average, recurrence occurs within five years following surgery in approximately half of the patients, regardless of the severity of their initial diagnosis [15]. This persistence and recurrence can be partly attributed to the presence of microscopic endometriotic lesions, which often remain undetected and can progress over time.

This multifaceted behavior of endometriosis underscores the need for more advanced diagnostic and therapeutic strategies to address its malignant-like characteristics and prevent complications effectively.

2. Case Presentation

A 45-year-old woman presented to the emergency room with severe abdominal pain that had only partially responded to ibuprofen during the previous two days. She also reported multiple episodes of vomiting and an inability to eat since the previous day. Her medical history was notable for stage IV endometriosis, surgically treated in 2008, and currently managed with estrogen–progestin therapy. The patient underwent the operation in a different hospital and no other information was available regarding the previous intervention. Additional comorbidities included fibromyalgia, successfully managed with muscle relaxants for 12 years, and pemphigus vulgaris, actually in remission following corticosteroid therapy. She also had a history of adenoidectomy during childhood.

Upon examination promptly after her ER admission, laboratory investigations revealed leukocytosis with a WBC count of $15,000/\mu$ L and anemia with a hemoglobin level of 9.8 g/dL. D-dimer levels were mildly elevated at 699 ng/mL, while other blood parameters were unremarkable. Imaging studies included a contrast-enhanced CT scan of the abdomen, which revealed significant hemoperitoneum, an enlarged and heterogeneous uterus, and evidence of contrast extravasation in the right para-uterine region, raising suspicion of a vascular rupture.

Given the clinical and radiological findings, the decision was made to perform an emergency laparotomy. Although the patient was awake and conscious, indeed, her BP was low (90/55) and her heartrate increased (149 BPM). Moreover, her Hb level was low. Laparotomy was chosen over laparoscopy due to the patient's hemodynamic instability and the urgent need for direct and rapid surgical intervention to control significant bleeding and hemoperitoneum. In emergencies with unclear sources of bleeding, laparotomy is often preferred for its comprehensive access and ability to address critical conditions

promptly. During surgery, approximately 600 cc of blood and clots was evacuated from the peritoneal cavity. The uterus appeared soft but of normal size, and the adnexa on the right were unremarkable. On the left side, a 5 cm hematoma involving the fallopian tube was identified. Further exploration revealed a rupture of the left paracervical vessels, which was successfully ligated to achieve hemostasis. Although the CT scan pointed out an extravasation in the left para-uterine region, raising the suspicion of a homolateral vascular rupture, the identified vessel was on the left. The authors attribute this to the hemoperitoneum. The peritoneal cavity was thoroughly lavaged, and no other sources of bleeding were identified. Since the patient had no active endometriotic lesions during the emergency laparotomy, the ASRM endometriosis scale and Enzian classification were not applicable in this case, as these scoring systems are used to assess the extent and severity of active endometriotic disease. During the emergency laparotomy, the ovaries were examined thoroughly and appeared unremarkable, with no visible changes or suspicious lesions. Given the absence of any areas suggestive of malignancy or pathology, no biopsies were taken for histopathological examination. Additionally, the clinical presentation and intraoperative findings did not raise concerns for ovarian cancer or metastatic disease. The focus was on addressing the vascular rupture and ensuring hemostasis, which was successfully achieved

Postoperatively, the patient's recovery was uneventful. She resumed normal bowel movements two days after surgery, and the surgical wound was intact. On discharge, she was afebrile, with stable vital signs and no evidence of complications, suggesting a favorable prognosis.

3. Discussion

A review of the literature was conducted to identify further cases of spontaneous rupture of the uterine arteries in non-pregnant patients with endometriosis. It is appropriate to exclude pregnant patients from this research as it is known that endometriosis represents an important risk factor for spontaneous hemoperitoneum in pregnancy, with a prevalence of this complication of 0.4% [16].

In Table 1, we observe the cases described in the literature from 1970 to 2024 regarding spontaneous vessel rupture in the case of endometriosis.

It is noteworthy that only 3 patients out of a total of 31 had a diagnosis of endometriosis before developing hemoperitoneum. In most of the cases described, the cause of the maximum bleeding is attributable to the rupture of endometriotic cysts located in the Douglas pouch or at the tubal, peritoneal, and sigmoid levels. Only three of the cases present in the literature and the clinical case proposed by us have as the cause of spontaneous hemoperitoneum a lesion affecting the uterine arteries.

This is a very rare complication of endometriosis. However, it is an acute clinical picture, and it is advisable to make an early diagnosis. This represents the main difference with the presented case, which reports a complication that occurred later on in life.

In these cases, the clinical picture is certainly decisive. We can use imaging techniques such as transvaginal ultrasound and CT to make a preoperative diagnosis and identify the source of bleeding. It is also appropriate to exploit the various imaging techniques available to make a differential diagnosis with hemorrhagic corpus luteum, ruptured adenomyotic uterus, or cause of hemoperitoneum detectable in the upper abdomen.

Author	Year	No. of Patients	Previous Diagnosis of Endometriosis	Treatment	Surgical Findings
Ranney B.	1970	4	not	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.
Kumar S.	1996	1	not	laparotomy	Active bleeding from endometriotic deposits in the pouch of Douglas and along both uterosacral ligaments.
Janicki et al.	2002	1	yes (stage III)	laparotomy	Left endometrioma (5 \times 6 cm); Erosion of the left uterine artery;
Evangelina	2009	16	not	laparoscopy	Ruptured left (n = 11) and right endometrioma (n = 5).
Togami et al.	2015	1	yes (site non specified)	laparoscopy	Active bleeding from endometriotic peritoneal deposits.
Lim et al.	2021	1	not	laparoscopy	Active bleeding from uterine artery erosion due to deep infiltrating endometriosis.

Table 1. Review of the literature.

In hemodynamically stable patients, it is possible to perform angiography and embolization [16]. In hemodynamically unstable patients, such as in our case, it is advisable to resort to surgery. From the table, we can see how, in recent years, the surgical technique used in this case is no longer laparotomy but laparoscopy, which is less traumatic for very fragile tissues and has an easier postoperative course for the patient.

Furthermore, when acute abdominal pain with hemoperitoneum occurs in nonpregnant women of reproductive age, in the absence of positive findings for liver or splenic lesions, a possible spontaneous rupture of the utero-ovarian vessels, although rare, related to the presence of previously unrecognized deep infiltrating endometriosis, should be included among the possible causes of the condition.

Spontaneous rupture of utero-ovarian vessels in non-pregnant women is an exceedingly rare but life-threatening event [17,18]. This case adds to the limited literature on such presentations, particularly in patients with a history of endometriosis. Notably, the patient had no active endometriotic lesions, emphasizing the unpredictability of vascular complications in chronic gynecological conditions.

A literature review identified only a few cases of spontaneous vascular rupture associated with endometriosis, with most attributed to the rupture of endometriotic cysts or erosion of uterine arteries by deep infiltrating endometriosis, with a different and acute presentation compared to the case we are presenting. In this case, the absence of active lesions underscores indeed the need for heightened clinical suspicion and advanced imaging to facilitate timely diagnosis.

The decision to proceed with laparotomy was dictated by the patient's hemodynamic instability and the need for direct surgical intervention. While less invasive approaches such as angiography and embolization are gaining popularity, laparotomy remains the gold standard in unstable patients or when the source of bleeding is unclear.

This case highlights several key considerations for clinicians:

- Differential diagnosis: In cases of acute abdominal pain with hemoperitoneum, spontaneous rupture of utero-ovarian vessels should be considered, particularly in patients with a history of endometriosis or other gynecological conditions.
- Role of imaging: Advanced imaging techniques, including transvaginal ultrasound and CT, are critical in identifying vascular injuries and differentiating them from other causes of hemoperitoneum.
- Multidisciplinary approach: Successful outcomes require prompt surgical intervention and collaboration among gynecologists, radiologists, and anesthesiologists.

In our case, the potential impact of fibromyalgia, muscle relaxants, or steroid use on the durability of the vessel is notable. While fibromyalgia is primarily a chronic pain condition, it is associated with systemic inflammation and altered tissue repair mechanisms, which might indirectly influence vascular integrity. Similarly, long-term use of muscle relaxants could hypothetically affect vascular tone, and chronic steroid use for pemphigus vulgaris might impair collagen synthesis and tissue healing, potentially weakening vessel walls. Although these factors were not discussed in detail in our report, they warrant further exploration as possible contributors to vascular rupture. Including such considerations in future discussions could help shed light on the multifactorial nature of this rare complication and its potential predisposing factors.

Further research is warranted to elucidate the mechanisms underlying spontaneous vascular rupture and to develop preventive strategies for at-risk populations.

This case report highlights several limitations. First, the condition described spontaneous rupture of utero-ovarian vessels in non-pregnant women—is exceedingly rare, making it difficult to generalize findings. The scarcity of reported cases limits the understanding of the underlying mechanisms and optimal management strategies. Second, diagnostic challenges arise due to the nonspecific presentation of symptoms, such as abdominal pain and hemoperitoneum, which overlap with other gynecological and non-gynecological conditions. Furthermore, the absence of active endometriotic lesions in the patient complicates the ability to establish a direct causal link between prior endometriosis and the vascular event. Finally, while imaging played a pivotal role in this case, its sensitivity in detecting spontaneous vascular ruptures remains variable, potentially delaying diagnosis and treatment in similar presentations [19,20].

4. Conclusions

This case underscores the rarity and clinical significance of spontaneous rupture of utero-ovarian vessels in non-pregnant women.

The rationale for presenting this case is to raise awareness of the rare but lifethreatening occurrence of spontaneous utero-ovarian vessel rupture, highlight diagnostic challenges, and emphasize the importance of timely surgical intervention for favorable outcomes. Moreover, the management protocol described could contribute to broader clinical guidelines for rare gynecological emergencies by providing a structured approach to diagnosing and managing spontaneous vascular ruptures, emphasizing the role of imaging, multidisciplinary collaboration, and timely surgical intervention.

Timely recognition and surgical intervention are paramount in managing such emergencies. Clinicians should maintain a high index of suspicion for this condition in patients presenting with acute abdominal pain and hemoperitoneum, particularly in those with a history of endometriosis. Advanced imaging and a multidisciplinary approach are essential for achieving favorable outcomes.

Further research should explore the mechanisms behind spontaneous vascular rupture in non-pregnant women, focusing on the role of chronic inflammation, hormonal influences, and vascular fragility associated with conditions like endometriosis. Additionally, studies could evaluate the effectiveness of advanced imaging techniques for early diagnosis and investigate preventive strategies in at-risk populations.

Author Contributions: Conceptualization, A.L.; methodology, V.R. and L.L.; validation, V.R.; formal analysis, A.L. and A.N.; investigation, A.L.; resources, A.L. and A.N.; data curation, V.R.; writing—original draft preparation, V.R., A.N. and A.L.; writing—review and editing, V.R. and A.L.; visualization, L.L.; supervision, V.R. and A.L.; project administration, L.L.; funding acquisition, V.R. All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Institutional Review Board Statement: The study was conducted in accordance with the Declaration of Helsinki. No Ethics Committee or institutional review board statement was required due to the nature of the case report (single patient who consented for publication). This is also in agreement with HIPAA (Health Insurance Portability and Accountability Act): "a case report is an activity to develop information to be shared for medical/educational purposes. Although the use of protected health information to prepare, the paper does not require IRB review".

Informed Consent Statement: Informed consent for participation was obtained from all subjects involved in the study.

Data Availability Statement: No new data were created or analyzed in this study. Data sharing is not applicable to this article.

Conflicts of Interest: The authors declare no conflicts of interest.

References

- 1. Zondervan, K.T.; Becker, C.M.; Missmer, S.A. Endometriosis. N. Engl. J. Med. 2020, 382, 1244–1256. [CrossRef] [PubMed]
- 2. Giudice, L.C. Clinical practice: Endometriosis. N. Engl. J. Med. 2010, 362, 2389–2402. [CrossRef] [PubMed]
- 3. Ballweg, M.L. Impact of endometriosis on women's health: Comparative historical data show that endometriosis is a common and serious disease that has significant impacts on all aspects of women's lives. *Am. J. Obstet. Gynecol.* **2006**, *195*, 565–572.
- 4. Bulun, S.E. Endometriosis. N. Engl. J. Med. 2009, 360, 268–279. [CrossRef] [PubMed]
- 5. Melis, I.; Vellino, L.; Nucera, D.; Marci, R. Chronic pain syndromes in women with endometriosis. *Int. J. Gynecol. Obstet.* **2016**, 134, 23–29.
- 6. Zondervan, K.T.; Becker, C.M.; Missmer, S.A. Fibromyalgia in patients with endometriosis. *Eur. J. Pain* 2019, 23, 150–156.
- Ferrero, S.; Ragni, N.; Remorgida, V. Migraine in women with endometriosis. J. Minim. Invasive Gynecol. 2005, 12, 534–539.
 [CrossRef]
- 8. Schindler, A.E. Endometriosis—The impact on migraine. *Climacteric* **2020**, *23*, 506–511.
- 9. Viganò, P.; Corti, L.; Berlanda, N. Beyond infertility: Obstetrical and postpartum complications associated with endometriosis. *Fertil. Steril.* 2015, 104, 802–812. [CrossRef] [PubMed]
- Kumar, S.; Khanna, S.; Roy, A.; Gupta, S.K. An unusual cause of hemoperitoneum: Case report with review of literature. *Int. J. Surg. Case Rep.* 2015, 12, 120–122. [CrossRef] [PubMed]
- 11. Fuldeore, M.J.; Soliman, A.M. Prevalence and symptomatic burden of diagnosed endometriosis in the United States: Evidence from a national survey. *Fertil. 2017*, *107*, 300–308.
- 12. Wheeler, J.M. Epidemiology of endometriosis-associated infertility. J. Reprod. Med. 1989, 34, 41–46. [PubMed]
- 13. Yeung, P.; Sinervo, K.; Winer, W.; Albee, R. Complete laparoscopic excision of endometriosis: Treating the root cause of the disease. *Fertil. Steril.* **2009**, *92*, 13–18.
- 14. Pandey, S. Metabolomics for the identification of biomarkers in endometriosis. *Arch. Gynecol. Obstet.* **2024**, *310*, 2823–2827. [CrossRef] [PubMed]
- 15. Hart, R.J.; Hickey, M.; Maouris, P.; Buckett, W. Excisional surgery versus ablative surgery for endometriosis. *Cochrane Database Syst. Rev.* **2005**, *4*, CD004992.
- 16. Berlanda, N.; Somigliana, E.; Vigano, P.; Vercellini, P. The impact of endometriosis on pregnancy. Hum. Reprod. 2010, 16, 1243–1253.
- 17. Tulandi, T.; Murray, C.; Guralnik, J. Laparoscopic treatment of endometriosis and pelvic pain. Obstet. Gynecol. 1998, 92, 944–949.
- Ferrero, S.; Bogliolo, S.; Rossi, U.G.; Baldi, C.; Remorgida, V. Unusual complication of excision of pelvic endometriosis: Pseudoaneurysm of the left uterine artery. *Fertil. Steril.* 2010, *93*, 264–266. [CrossRef] [PubMed]

- Centini, G.; Schettini, G.; Pieri, E.; Giorgi, M.; Lazzeri, L.; Martire, F.G.; Mancini, V.; Raimondo, D.; Seracchioli, R.; Habib, N.; et al. Endometriosis-Related Ovarian Cancer: Where Are We Now? A Narrative Review towards a Pragmatic Approach. *J. Clin. Med.* 2024, 13, 1933. [CrossRef] [PubMed]
- 20. Libretti, A.; Dealberti, D.; Prigione, S.; Remorgida, V. It's never endometriosis, except when it's endometriosis. *Acta Bio-Med. Atenei Parm.* **2023**, *94*, e2023010. [CrossRef]

Disclaimer/Publisher's Note: The statements, opinions and data contained in all publications are solely those of the individual author(s) and contributor(s) and not of MDPI and/or the editor(s). MDPI and/or the editor(s) disclaim responsibility for any injury to people or property resulting from any ideas, methods, instructions or products referred to in the content.