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Case report

Spontaneously ruptured endometriomas presenting with symptoms and imaging findings worrisome for carcinomatosis: A case report

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ABSTRACT

Introduction and importance: Endometriomas are the most common presenting subtype of endometriosis. Although most endometriomas are asymptomatic, patients can rarely present acutely with spontaneous rupture causing diffuse peritonitis and severe systemic inflammatory response.

Case presentation: Here we describe a case of ruptured endometriomas in a 26-year-old nulligravid female with a history of heavy menses, progressive abdominal distension, and a recent urinary tract infection. The patient presented to the emergency department with upper abdominal pain radiating to her back with associated nausea. Computed tomography (CT) scan demonstrated diffuse ascites with a large, multilobulated, and multicystic septated mass arising in the right pelvis and extending into the lower abdomen. Findings were concerning for peritoneal carcinomatosis and the patient was admitted for evaluation. She developed progressive signs of sepsis and was emergently brought to the operating room for surgical exploration on hospital day (HD) number two. She was found to have ruptured pelvic cysts arising from both ovaries with diffuse contamination of the abdomen by cyst contents and bilateral salpingo-oophorectomy (BSO) was performed. Final pathology demonstrated benign bilateral endometriomas.

Clinical discussion: Endometrioma rupture is extremely rare and imaging findings may appear to represent disseminated peritoneal malignancy. CT findings demonstrating a pelvic mass with concurrent ascites should raise clinical suspicion for ruptured endometrioma, particularly in younger patients.

Conclusion: Prompt surgical exploration and complete resection of pathologic tissue may be necessary for diagnosis and treatment in some patients with clinical deterioration related to perforated endometriomas. Combined oral contraceptives are recommended in the postoperative period.

1. Introduction

Endometriosis is a chronic inflammatory disease of extra-uterine endometrial tissue that affects approximately 10 % of women of reproductive age. Endometriomas, or endometriotic ovarian cysts, arising in the ovary are the most common manifestation. Endometriomas are generally asymptomatic early in their development, but patients may develop chronic pelvic pain, dyspareunia, dysmenorrhea, and dyschezia as the lesions grow [1]. Prolonged estrogen exposure has been shown to increase the risk of development [2]. Although rare, spontaneously ruptured endometriomas can present with acute abdominal pain, fever, nausea, vomiting, and peritonitis [3]. Transvaginal ultrasonography (TVUS) and magnetic resonance imaging (MRI) are the most accurate imaging studies used to evaluate suspected cases [1]. The gold standard for diagnosis and treatment of a ruptured or non-ruptured endometrioma is achieved by diagnostic laparoscopy [3–5], although surgical exploration may be necessary in cases of clinical deterioration or diagnostic uncertainty [3,5–10] similar to our own. Cystectomy is another treatment option for endometriomas, however there is a higher risk of complications and recurrence of endometriomas [4,5]. Definitive treatment is achieved by total abdominal hysterectomy (TAH) with bilateral salpingo-oophorectomy (BSO) [3].

Here we present a case of a ruptured endometrioma causing hemorrhage, chemical peritonitis, and clinical deterioration in a patient presenting with abdominal pain and CT imaging demonstrating cystic pelvic masses and diffuse ascites. SCARE criteria [11] were followed

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2. Presentation of case

A 26-year-old nulligravid woman presented to the emergency department with nausea and upper abdominal pain radiating to her back. She endorsed a five-day history of vaginal bleeding since her last menstrual period. Past medical history was significant for asthma, heavy menses, progressive abdominal distension, and a recent urinary tract infection. She was treated with amoxicillin for an upper respiratory infection two days prior to presentation. She had no past surgical history. She was sexually active in a monogamous relationship and was not using any form of birth control. She had not routinely seen a gynecologist. Family history was notable for colon cancer in her mother who died at the age of 50. The remaining medical history was unremarkable.

On physical examination, the patient had a temperature of 38.6 °C (101.5 °F) and was tachycardic with a heart rate of 124. Her body mass index (BMI) was 29.5. Her abdomen was distended, but soft. There was some tenderness in the right upper quadrant, but there were no peritoneal signs. Diagnostic laboratory testing showed a white blood cell (WBC) count of 16.4 $\times$ 10^9/L and a hemoglobin level of 9.4 g/dL. Ultrasound (US) confirmed four-quadrant ascites. Abdominal and pelvic CT scan with contrast demonstrated diffuse ascites with a large, multilobulated, and multicystic septated mass arising from the right pelvis, extending into the lower abdomen, and measuring approximately 16 cm in maximal dimension (Fig. 1).

There was concern for an advanced ovarian neoplasm based on the CT findings and her indolent clinical presentation. In light of her signs of sepsis, she was empirically treated with broad spectrum antibiotics. Tumor markers were notable for CA-125 of 3741 U/mL, LDH of 550 U/L and CEA of <0.5 ng/mL. Blood cultures were negative. Qualitative serum beta-hCG was negative. Cervical amplified probe test and urine testing were both negative for chlamydia, gonorrhea, and trichomonas.

The patient was admitted to the hospital for stabilization and planned referral to a specialist in gynecological oncology. General surgical consultation was requested, and recommendations were made for paracentesis with analysis of peritoneal fluid and consideration for diagnostic laparoscopy. The patient experienced a continued decline in her clinical status over the next 48 h showing worsening tachycardia with a heart rate of 144, despite aggressive volume replacement, and leukocytosis with a WBC of 30 $\times$ 10^9/L. She was noted to have progressive abdominal distention and tenderness, shortness of breath, and an oxygen saturation in the 80s. CT of the chest, abdomen, and pelvis demonstrated bilateral lower lobe consolidation and pleural effusions consistent with acute respiratory distress syndrome (ARDS) and extensive ascites with a large, multiseptated mass arising in the right pelvis, extending into the lower abdomen, and measuring approximately 16 cm in maximal dimension. Due to her progressive clinical deterioration, concerns for an intra-abdominal source, and diagnostic uncertainty, the patient was brought to the operating room for emergent surgical exploration. Diagnostic laparoscopy was attempted, but visualization was extremely poor, and the procedure was converted to open laparotomy. Intraoperative abdominal findings were notable for large volume of brown fluid and debris distributed throughout the peritoneal cavity

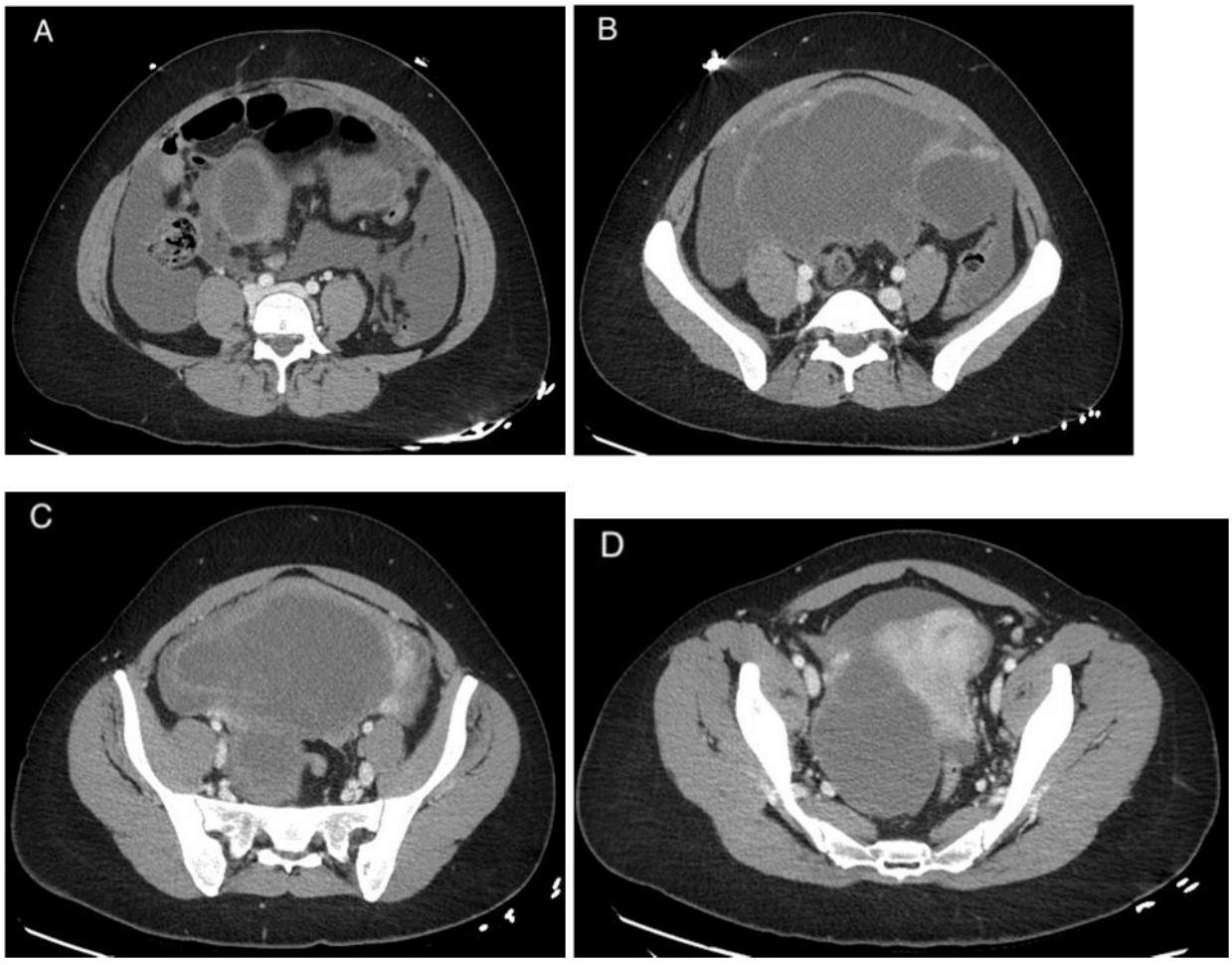

Fig. 1. Transverse abdominal CT scan (A-D) shows extensive ascites and a large multiseptated, multicystic mass arising in the right pelvis and extending into the lower abdomen, measuring approximately 16 cm in maximal dimension.

with diffuse peritonitis. A large right-sided cystic pelvic mass and two additional thick white/tan cystic masses attached to each ovary were identified. The larger of the two additional masses was arising from the left ovary and had a posterior perforation with notable spillage of the contents. The left ovary was enlarged with additional smaller masses including a simple cyst. The right ovarian cystic lesion was thickened, blue/purple in color, without torsion, adherent to the appendix, and noted to be ruptured as well. Complete resection of both cystic lesions was performed and required *en bloc* appendectomy and BSO. Omentectomy was performed for possible staging. Free abdominal fluid and debris were aspirated and evacuated, and the abdomen was copiously irrigated with normal saline. There were no endometriosis implants found in the abdomen. The surgical team elected to leave the abdomen open with plans for continued resuscitation in the intensive care unit (ICU) and return to the operating room for a second look laparotomy in 24–48 h. The abdomen was managed with ABThera™ (KCI USA Inc., San Antonio, TX) negative pressure therapy. Estimated blood loss was 50 mL.

The patient remained intubated postoperatively and was transferred to the ICU for aggressive resuscitation and monitoring. Intraoperative peritoneal cultures showed no growth. On postoperative day (POD) 1, the patient's WBC count dropped to 12.2×10^9/L. The patient received transfusion with two units of packed red blood cells (PRBC) for a declining hemoglobin of 6.7 mg/dL prior returning to the operating room for formal closure on POD 2. Peritoneal surfaces appeared to be chronically inflamed at time of surgical re-exploration, but there was no evidence of untreated infection or hemorrhage. These findings suggested her anemia was related to blood loss occurring at the time of cyst rupture. The abdomen was closed with interrupted nonabsorbable sutures and retention sutures with bolsters. The patient recovered uneventfully and was discharged to home on POD 8. The patient was readmitted to the hospital approximately two weeks after her surgery for colonic distension that was treated with bowel rest and IV fluids. CT of the abdomen and pelvis done at that time showed only postsurgical changes with no re-accumulation of intra-abdominal fluid or other abnormalities. She was started on hormonal replacement therapy with a combined estradiol transdermal patch and progesterone pill by her gynecologist and resumed her normal activities.

Final pathology was remarkable for benign bilateral endometriomas measuring at $8 \times 4.8 \times 2.1$ cm and $14 \times 9.3 \times 2.4$ cm in size. The omentum was found to have no pathologic changes. Histologic and immunohistochemical analysis of the ovarian cysts were positive for endometrial glandular epithelium, endometrial stroma, and areas of hemosiderin laden macrophages. This constellation of findings supported a diagnosis of endometriosis, and since they were large cystic structures, endometriomas (Fig. 2).

3. Discussion

Endometriosis is defined as an inflammatory disease of

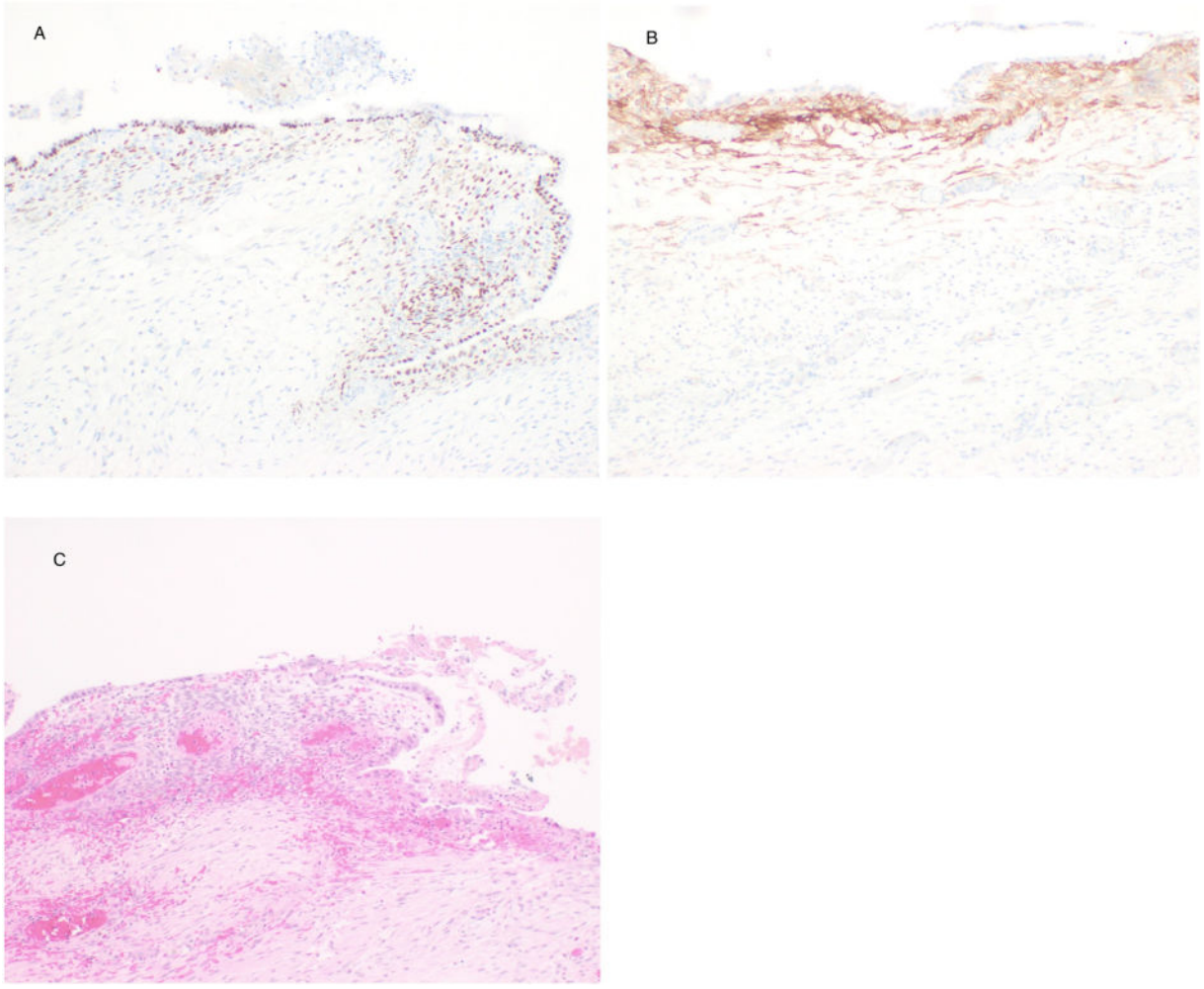

Fig. 2. (A) ER stains positive for endometrial glandular epithelium, (B) CD-10 stains positive for underlying endometrial stroma, but negative for the overlying endometrial epithelium and the underlying ovarian stroma, (C) H&E stains positive for epithelial layer on top, supporting endometrial stroma layer, and native ovarian tissue.

endometrium-like epithelium and/or stroma outside the endometrium and myometrium [12]. Endometriosis commonly presents as peritoneal lesions, deep endometriosis, or endometriomas, and these can frequently occur concurrently [1]. Approximately 17–44 % of patients with endometriosis are known to have endometriomas [2]. Endometriomas, or ovarian endometriotic cyst, are defined as endometrium-like tissue in the form of ovarian cysts. These cysts usually contain dark, blood-stained fluid and commonly are referred to as "chocolate cysts" [12]. It is widely believed endometriomas develop from a combination of either retrograde menstruation or celomic metaplasia of ovarian epithelium [3]. Associated risk factors include nulliparity, early menarche, and heavy menstrual bleeding. With prolonged exposure to estrogen, endometrioma growth responds through proliferation, secretion, and shedding. The sloughing of endometrial tissue and subsequent formation of fibrous tissue may lead to chronic abdominal or pelvic pain [2]. <3 % of patients with endometriosis experience a spontaneously ruptured ovarian endometrioma [6]. Moreover, it is exceedingly rare to have the simultaneous occurrence of endometriosis and ascites [10,13].

Within the existing literature, it is more common, however rare, for patients to present with ruptured endometriomas during pregnancy and after abdominal or pelvic trauma [6]. Endometriomas often present in an indolent fashion depending on size and extrinsic forces. Symptoms can progress to dysmenorrhea, abdominal or pelvic pain, dyspareunia, dyschezia, and in some cases, rupture can occur [6,7,9]. Patients may present acutely with fever, nausea, vomiting, rebound tenderness, guarding, and peritonitis [8]. Here we report the case of a spontaneously ruptured, noninfected endometriomas presenting with CT imaging worrisome for carcinomatosis in a premenopausal, nulligravid patient. Serum cancer antigen 125 (CA-125) testing can be useful for establishing the diagnosis of ovarian cancer, monitoring response to treatment, and surveillance for identifying disease recurrence [14]. Notably, elevated CA-125 values can be nonspecifically elevated in patients with ruptured endometriomas [15–17]. Furthermore, recent studies have shown no difference between preoperative CA-125 levels in patients with ovarian neoplasms and ovarian endometriomas [14]. Consequently, CA-125 does not appear to be useful for differentiating endometriomas from ovarian cancer.

The patient's history of abdominal issues, heavy menses, and lack of birth control may have contributed to her development of endometriomas. TVUS is the recommended initial imaging test of choice for endometrioma with reported sensitivities and specificities >90 % [1]. If ultrasound is inconclusive, CT or MRI imaging are acceptable alternatives, and can offer additional information regarding relationship of lesions to adjacent structures. Numerous salient features of this case such as progressive abdominal pain and distention, fever, worsening tachycardia, and imaging findings of a pelvic mass with concurrent ascites were suggestive of ruptured endometrial cysts. However, ruptured dysgerminomas, torsion of adnexa, Meigs syndrome, or primary peritoneal tumors can present in a similar manner [13]. Although there was no evidence of bacterial peritonitis on culture, spillage of intralesional contents appears to have resulted in chemical peritonitis. Endometriosis-related ascites and subsequent peritonitis has been theorized to result from the spontaneous rupture of endometriotic cysts which produce reactive peritoneal fluid [13]. This aseptic inflammatory peritoneal reaction can result in complications such as adhesions, abdominal wall abscess, or enterocutaneous fistulas resulting in successive abdominal surgeries [8,9].

Management strategies for endometriosis are based on patient-related factors such as clinical presentation, extent of disease, and desire for fertility. Laparoscopic removal of endometriomas is generally recommended in an elective setting since drainage alone leads to a high recurrence rate [3]. Cystectomy can result in ovarian tissue damage and subsequent fertility issues. Ovarian cystectomy using the stripping or ablation technique can be used in selected patients in order to preserve fertility [4], although there is a risk of recurrent endometrial cysts [5], and a reduction in ovarian reserve measured by a decrease in anti-Mullerian hormone (AMH) [4]. Studies have shown the severity of endometriosis with adhesions involving the ovaries and/or fallopian tubes or bilateral endometriomas correlates with an increased risk of recurrence postoperatively [3,5]. Indications for definitive surgical treatment by TAH with BSO include failure of more conservative medical and surgical management, desire for infertility, or acute clinical decompensation [3,8–10,13]. In the described case, the constellation of cyst size, relation to the ovarian tissue, clinical status, and diagnostic uncertainty mandated a more aggressive management strategy despite the patient's young age. In general, the available literature on endometriosis-related ascites is limited to case reports and case series. As such, no definitive statements regarding optimal management can be made at this time [13].

Regardless of surgical treatment, all patients should be placed on hormone replacement therapy in order to alleviate pain and prevent recurrence. The goal of pharmacotherapy in the postoperative period is to suppress ovarian function and promote complete resolution of any residual endometriotic ovarian cyst. Continuous combined oral contraceptive (COC) has been shown to be the most effective relative to a cyclic COC, gonadotropin-releasing hormone (GnRH) agonists, or progesterone-releasing intrauterine systems [3–5].

4. Conclusion

Endometrioma rupture is rare and can present with nonspecific symptoms that can rapidly progress and cause clinical deterioration. Imaging findings of a pelvic mass with concurrent ascites in patients with abdominal complaints and signs of sepsis should raise clinical suspicion, especially in patients with risk factors such as pregnancy, young age, or history of trauma. Although these findings can suggest an advanced intra-abdominal or peritoneal malignancy, clinical deterioration should prompt urgent surgical exploration. Complete resection of pathologic tissue may be necessary for diagnosis and treatment in some patients with perforated endometriomas. Combined oral contraceptives are recommended in the postoperative period to manage pain and prevent recurrence of endometriotic ovarian cysts.

Ethical approval

This case report got ethical approval from our institution. The patient was given the written consent form.

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Credit authorship contribution statement

Connor Foote provided substantial contributions to acquisition of data and drafted the article; Peter Muscarella, MD, Hannah Bailey, MD, MD, Wilfrido D. Mojica, MD, Amber Milward, MD, and Alexa D. Smith revised the article critically for important intellectual content; Peter Muscarella, MD gave final approval of the version of the article to be published; Wilfrido D. Mojica provided histopathological images and data; and all authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Guarantor

Peter Muscarella

Research registration number

Not applicable.

Informed consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

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Declaration of competing interest

None declared.

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