



Rare Broad Ligament Cavernous Hemangioma Mimicking Advanced Endometriosis: A Case Report

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Abstract

Cavernous hemangioma within the female genital tract is an extremely rare pathology, characterized by irregular vascular spaces containing blood or thrombus. We present a unique case of a 42-year-old primiparous woman who presented with typical endometriosis symptoms such as dysmenorrhea, dyspareunia, and heavy menstrual bleeding. The patient also experienced complex postpartum symptoms, which were misdiagnosed as cholecystitis and retained placental products. Imaging studies suggested deep infiltrative endometriosis with extraovarian endometriotic lesions. Surgical exploration revealed a hemangioma within the right anterior broad ligament alongside peritoneal endometriosis lesions. The hemangioma itself expresses estrogen and progesterone receptors in stromal cells. The presence of steroid hormone receptors strongly suggests symptom alleviation during the menstrual cycle and the postpartum period. The coexistence of cavernous hemangioma and endometriosis in the broad ligament, previously unreported, and symptomatic overlap between the two conditions complicates diagnosis and management, emphasizing the need for comprehensive evaluation integrating clinical symptoms and imaging findings.

Keywords Cavernous hemangioma · Endometriosis · Dysmenorrhea · Dyspareunia · Menorrhagia

Introduction

Cavernous hemangiomas are vascular tumors that are extremely rare within the genital tract and characterized by irregular anastomosing vascular spaces lined with endothelial cells and containing intraluminal blood or thrombus [1]. Cavernous hemangiomas have been documented in 11 case reports in the fallopian tubes and 60 cases in the ovaries, presenting with symptoms such as abnormal uterine bleeding, postpartum hemorrhage, dyspareunia, and postcoital

bleeding. However, they have not yet been reported in the broad ligament [2–4]. Despite the advancements in understanding these tumors, the pathophysiology of cavernous hemangiomas has not been fully understood. It has been observed that estrogen contributes to their development by increasing the number of endothelial progenitor cells and various angiogenic factors [5]. Hormonal changes during pregnancy have been suggested as a potential alleviating factor in the growth and proliferation of cavernous hemangiomas [6].

Endometriosis is a chronic inflammatory condition characterized by functional endometrial-like glands and stroma outside the uterus. It affects approximately 10–15% of women of reproductive age, with the pelvic region being the most frequent site of occurrence [7]. Common symptoms of endometriosis include dysmenorrhea, dyspareunia, lower abdominal pain, and pelvic pain. Moreover, dyschezia, lower back pain, and dysuria are frequently reported findings [8].

Hereby, we present the case of a cavernous hemangioma within the right broad ligament, coexisting with symptomatic peritoneal endometriosis lesions.

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CASE

A 42-year-old primipara patient was admitted due to endometriosis evaluation due to complaints of dysmenorrhea, dyspareunia, and heavy menstrual bleeding. A patient also reported experiencing chronic right-sided sharp leg pain, diarrhea, and bloating during her menstrual cycles. Previously, an oral contraceptive pill containing ethinylestradiol and progestin was prescribed to a patient, resulting in a period free of symptoms from puberty until her pregnancy. However, after delivery, a patient preferred not to undergo medical treatment because of a family history of breast cancer.

The patient experienced prolonged vaginal bleeding lasting for 13 months after an uncomplicated pregnancy. Dilation and curettage was performed 6 weeks post-delivery and yielded normal results. She also endured severe gastrointestinal (GI) symptoms, such as vomiting, heartburn, and cramping, during the postpartum period. Her GI symptoms were attributed to cholecystitis, and cholecystectomy was performed 6 weeks post-delivery; however, pathology results were normal.

The initial assessment of the patient, including transvaginal ultrasound, revealed a retroverted uterus with increased blood flow, a hydrosalpinx in the right adnexa, and normal bilateral ovaries. Magnetic resonance imaging (MRI) evaluation showed mild thickening along the posterior uterine

body and pelvic ligaments, displaying a T2 hypointense signal associated with deep pelvic endometriosis. A distended tubular-like multiloculated cystic lesion was also observed in the right adnexa (Fig. 1). The main preoperative diagnoses included concurrent tubal pathologies or an extraovarian endometriotic lesion along with deep endometriosis.

During surgery, an unexpected finding was a coexisting pink-purple, blueberry-like cystic mass at the right anterior broad ligament instead of hydrosalpinx on MRI (Fig. 2). The histopathological evaluation of the excised specimen from the right broad ligament revealed a cavernous hemangioma, with CD31 positivity observed on immunohistochemistry. Notably, stromal cells stained positive for estrogen receptor (ER) (patchy weak) and progesterone receptor (PR) (diffuse strong), indicating hormonal receptor positivity in the hemangioma (Fig. 3).

The patient was diagnosed with stage 2 endometriosis according to ASRM, with no advancing endometriosis. Endometriotic implants were visualized by the aqua-blue contrast technique [9] in the left cul-de-sac and posterior cervix (Fig. 4). During surgery, evaluation of the peritoneum to ensure complete removal of endometriosis lesions is vital for effective symptomatic treatment, the aqua-blue contrast technique increased the peritoneal contrast and potentially enhanced lesion detection.

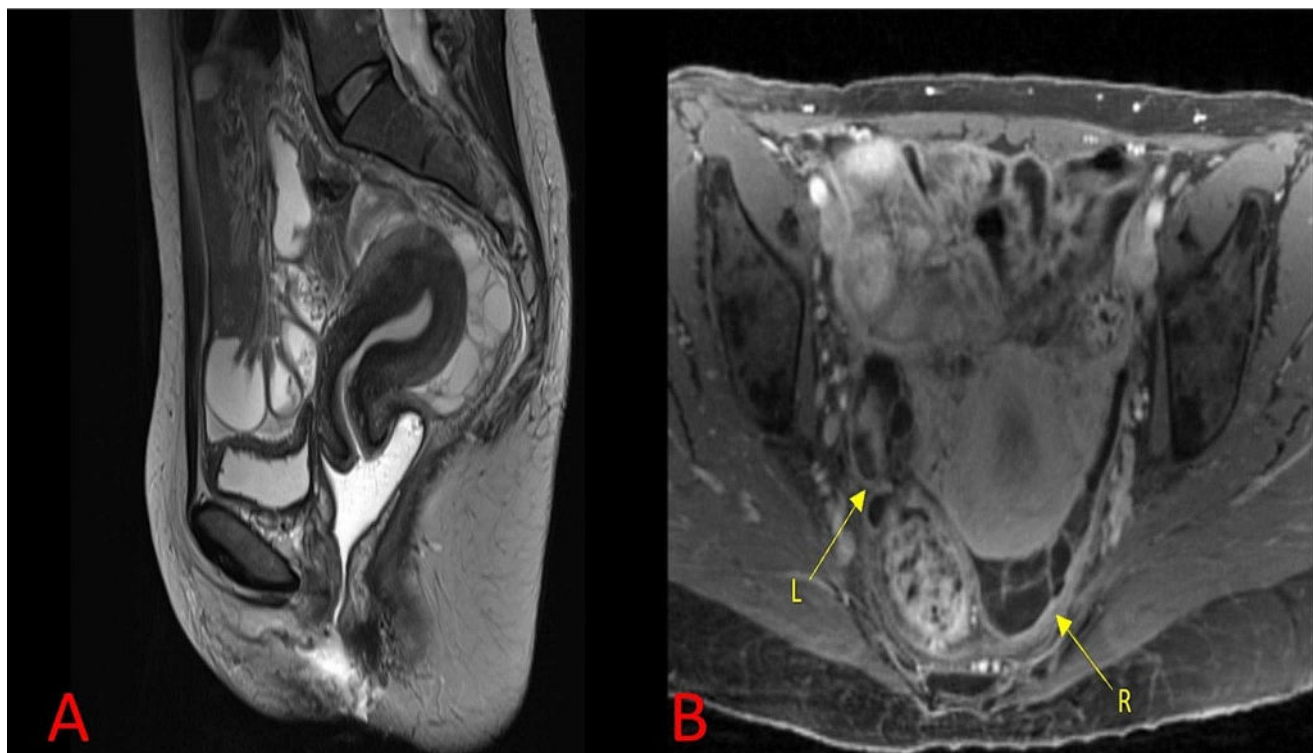


Fig. 1 MRI findings. MRI: magnetic resonance imaging. R: right adnexa L: left adnexa. **A:** Mild thickening along the posterior uterine body and pelvic ligaments (T2 signal sequence). **B:** Cystic mass in the right adnexa (T1 signal sequence)

Fig. 2 Cavernous hemangioma at the right anterior broad ligament (1.9×1.7×1.0 cm). Arrow shows the cavernous hemangioma lesion. Blue marking shows right round ligament

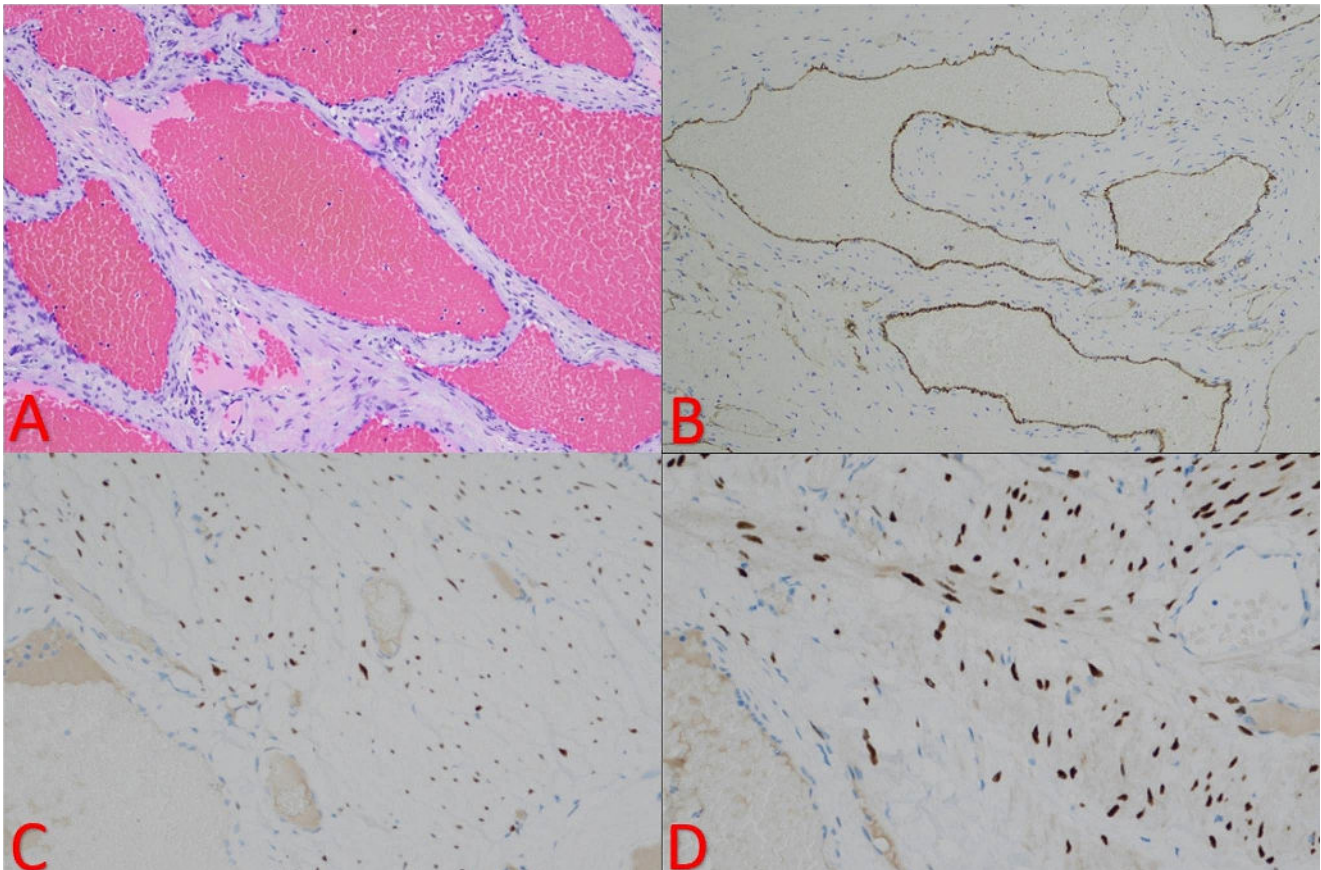
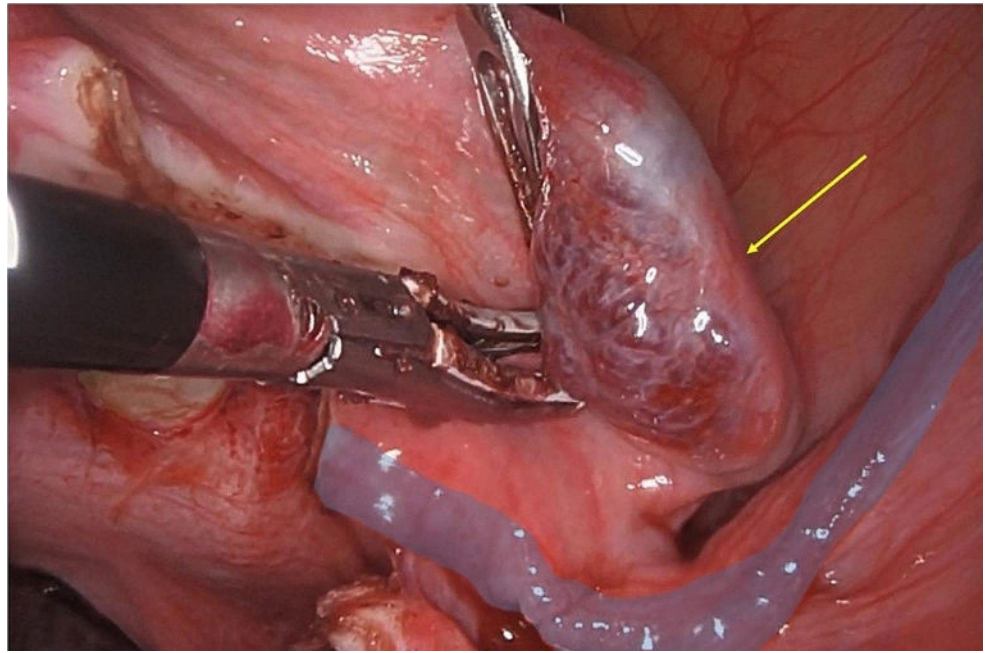


Fig. 3 Histopathological evaluation of the cavernous hemangioma at the right anterior broad ligament. H&E: hematoxylin eosin, PR: progesterone receptor, ER: estrogen receptor. **A:** 100x magnification H&E: spaces lined by single layer of endothelium. **B:** CD31-200X- the

positive endothelial lining of vascular spaces. **C:** ER- 200X- weak focal positivity in intervening stroma. **D:** PR- 200X- strong positivity in intervening stroma

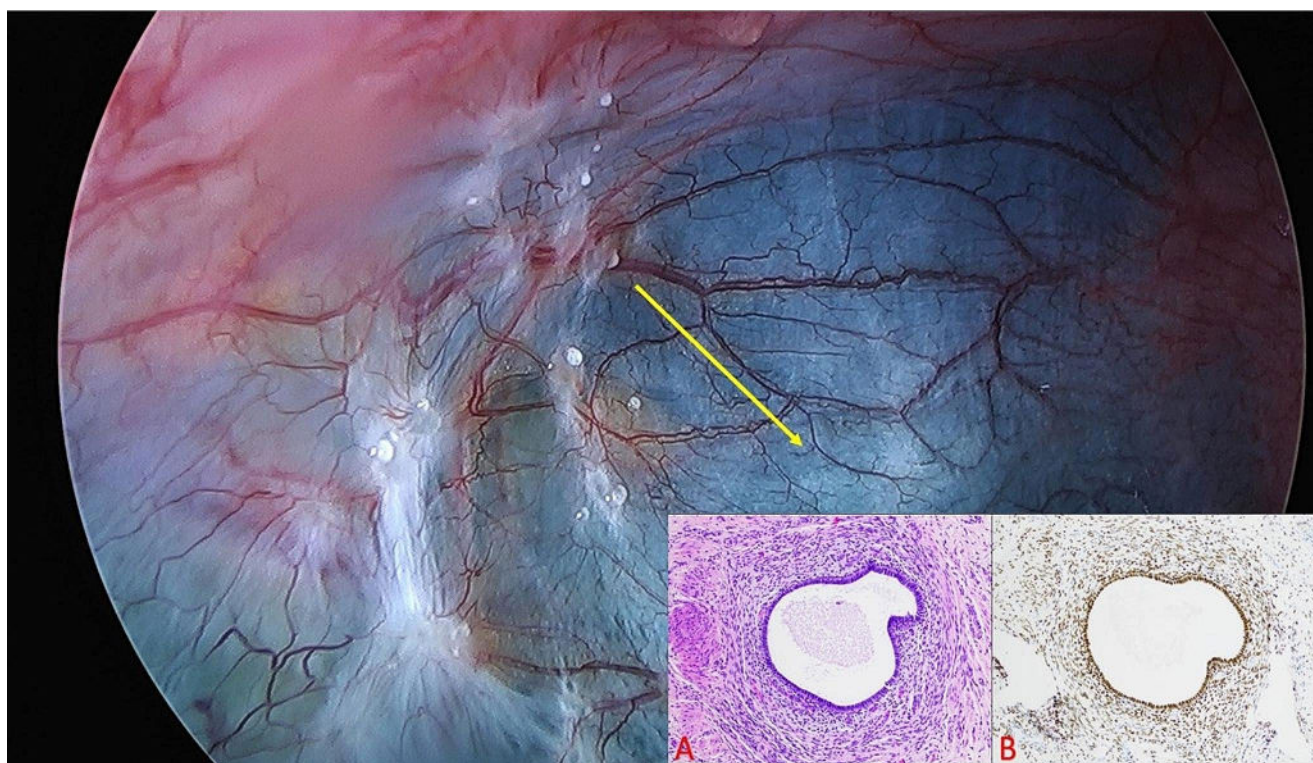


Fig. 4 Endometriotic implants at the peritoneum. The vascularization and thickening of the peritoneum around the left cul-de-sac endometriosis lesion were observed following the insertion of aqua-blue dye

(1.5×1.0×0.2 cm). H&E: hematoxylin eosin. ER: estrogen receptor. **A:** 100x magnification H&E: endometriosis. **B:** ER– 100X– positivity in glands and stroma

Discussion

The occurrence of hemangioma in the broad ligament of the genital tract has not previously been reported, and a noteworthy finding, with the exact pathology remains unclear. Acquired forms are more commonly linked to hormonal and physiological changes [10]. Imaging techniques like ultrasound and MRI can help distinguish hemangiomas, but definitive diagnosis relies on surgical removal of the lesion followed by histopathological evaluation [11].

Previous experimental and clinical studies have suggested estrogen and progesterone may induce hemangioma development by affecting the angiogenesis process [12]. ER expression has been detected in the endothelial cells and stroma of hemangiomas in the reproductive tract [13]. PR is important for the growth of hormone-sensitive fibroid-like smooth muscle lesions and its expression changes with ER in the endometrium during the menstrual cycle [14]. High postpartum steroid hormone levels and positive receptor expression in the affected area strongly correlate with worsened clinical symptoms, including prolonged vaginal bleeding and GI issues. Moreover, defective estradiol production on the endometriotic tissue and amplified steroid hormone response flare up the endometriosis symptoms

[15]. Symptoms could have originated from endometriosis itself or were influenced by the presence of a hemangioma.

Endometriosis has three anatomical subtypes: peritoneal, ovarian, and deep infiltrative. Peritoneal endometriosis is described as superficial darkly pigmented lesions resembling “powder burns”, however, non-pigmented vesicular lesions are also highly common [16]. When peritoneal endometriosis is seen during the surgery, it is essential to remove the endometriotic tissue due to the poor correlation between the severity of the endometriosis, and lesion subtypes [17].

Although endometrioma is described as endometriosis in the ovaries, endometriomas can rarely be present outside the ovarian tissue. Extraovarian endometriomas can manifest independently in cases of pelvic pain without peritoneal or ovarian involvement and/or adhesive disease, and only a few cases have been reported in the broad ligament with a multicystic appearance [18]. In this case, we evaluated this as a potential diagnosis preoperatively based on the imaging findings and the atypical clinical complaints described by the patient. One should maintain suspicion of extraovarian endometrioma in patients with atypical cystic masses and normal ovaries, particularly suspicion of endometriosis.

MRI’s effectiveness in identifying peritoneal endometriosis is limited, with sensitivity, specificity, accuracy, and PPV of 14%, 76%, 70%, and 7%, respectively, as reported

[19]. Indeed, its low sensitivity may result in the oversight of smaller peritoneal and non-ovarian lesions. Before surgical intervention, symptoms and MRI strongly suggested deeply infiltrative endometriosis with a cystic adnexal lesion. However, laparoscopic assessment revealed a different clinical perspective. It is important to approach endometriosis cases by evaluating their clinical manifestations, and one should also be mindful that peritoneal lesions have an important role in the pain profile.

Conclusion

This case presents an uncommon cooccurrence of ER and PR expressing cavernous hemangioma within the broad ligament and symptomatic peritoneal endometriosis, emphasizing the diagnostic and management challenges, and the significance of integrating clinical symptoms and imaging findings. Moreover, the concurrent presence of hemangiomas and endometriosis in the pelvic region has not been reported.

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Code Availability Not applicable.

Declarations

Ethics Approval Not applicable.

Consent to Participate Informed consent is taken from the patient for participation and publication.

Consent for Publication Informed consent is taken from the patient for participation and publication.

Competing Interests The authors declare that they have no competing interests. Timur Seckin, Hakan Kula, Amanda Chu, and Sabina Hajiyeva declare they have nothing to disclose. Tamer Seckin declares that he has a leadership role with the Endometriosis Foundation of America.

Attestation Statement Data regarding any of the subjects in the study has not been previously published unless specified. Data will be made

available to the journal editors for review or query upon request.

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