



CASE REPORT

# Thrombosis of iliac vessels, a rare complication of endometriosis: Case report and review of literature



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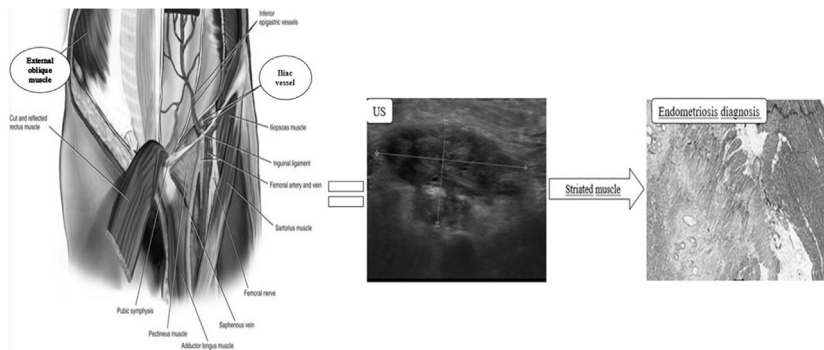
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GRAPHICAL ABSTRACT



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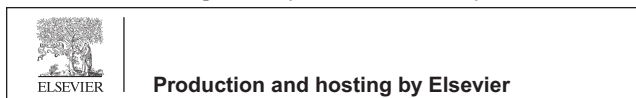
ABSTRACT

A young women presented with iliac vein thrombosis, as extrapelvic endometriosis complication. Endometriosis mass had a subfascial position at the level of external oblique muscle and extended to the iliac vein. This paper reviewed the literature on endometriosis cases localized into striated muscles and cases of deep vein thrombosis due to this disease. There are not similar cases in the literature. The diagnostic role of ultrasound, in obtaining the definitive

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histological diagnosis, may be further enhanced through Elasticity Imaging Techniques and ultrasound-guided biopsy.

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## Introduction

Endometriosis is an estrogen-dependent inflammatory process. Cyclic hormonal changes produce different symptoms depending on the site of implantation. Pelvic endometriosis primarily affects ovaries, pelvic peritoneum, utero-sacral ligaments, Douglas pouch, vagina, rectum and bladder. But different rarer localizations are described. The history of present illness and an accurate assessment through Ultrasound (US) examination leads physician to a correct diagnosis, management and follow-up. But an extraperitoneal localization makes radiologic imaging diagnosis more difficult.

The aim of this study was to assess the precision of US scanning in identifying the specific features of endometriosis and its eventual associated complications. The study illustrated the precision and the appropriateness of ultrasound-guided biopsy in providing definitive histological diagnosis of this rare condition.

This paper reviewed endometriosis cases with regard to importance of US examination for the diagnosis of the disease localized into striated muscles and for the follow-up of deep vein thrombosis (DVT) as its complication. Cases of endometriosis localized in external oblique muscle and associated with DVT have not been previously reported in the literature.

## Case report

*All procedures performed in our study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from the participant included in the study.*

A 38-year-old white woman, with no previous pregnancies, presented to hospital for acute onset of right groin pain. She described her menses as painless and regular. She denied episodes of menorrhagia, metrorrhagia, dyspareunia, dysmenorrhea, abdominal and pelvic cramping.

The lower limb appeared swollen warm and with an erythematous skin rash. Pulses, sensation and strength were intact. The symptomatology did not seem associated with menstruation and the patient did not report similar previous events.

## Results

Ultrasound examination, using a convex multi-frequency probe with a range of 1.0–6.0 MHz, showed normal morphological structures of abdomen. A solid fusiform mass, with a

diameter of  $4.45 \times 2.18$  cm, was localized in the right inguinal region, near to the iliac vessels. The mass had a sub-fascial localization in the right external oblique muscle, adjacent to subcutaneous fat. It had an inhomogeneous echotexture with low level internal iso-hypochoic echoes, irregular borders, without an endometrioid aspect (Fig. 1a). It was very extensive and compressed the iliac vein, without evidence of DVT during the first evaluation. Doppler ultrasound showed a diffuse peripheral vascularization (Fig. 1b). The shear wave elastographic evaluation (Elasticity Imaging Techniques) showed a higher elasticity in the peripheral region of the lesion. The central part of the lesion presented a homogeneous blue color distribution, representing hard stiffness (Fig. 1c). Computed tomography, with intravenous contrast, revealed a 22 mm irregular thickening, with spiculated margins, located in the subfascial of external oblique muscle (Fig. 2a). It was thickened and inhomogeneous. Regional lymphadenopathy was present.

Differential diagnosis included hematoma, abscess, enlarged lymph node, desmoid tumor and malignant tumors such as malignant melanoma, sarcoma or metastatic carcinoma. Percutaneous biopsy, using a 16 gauge Tru-cut needle system, was performed under US guidance. Histopathological examination confirmed endometriosis nodule involving the muscular layer of the external oblique muscle.

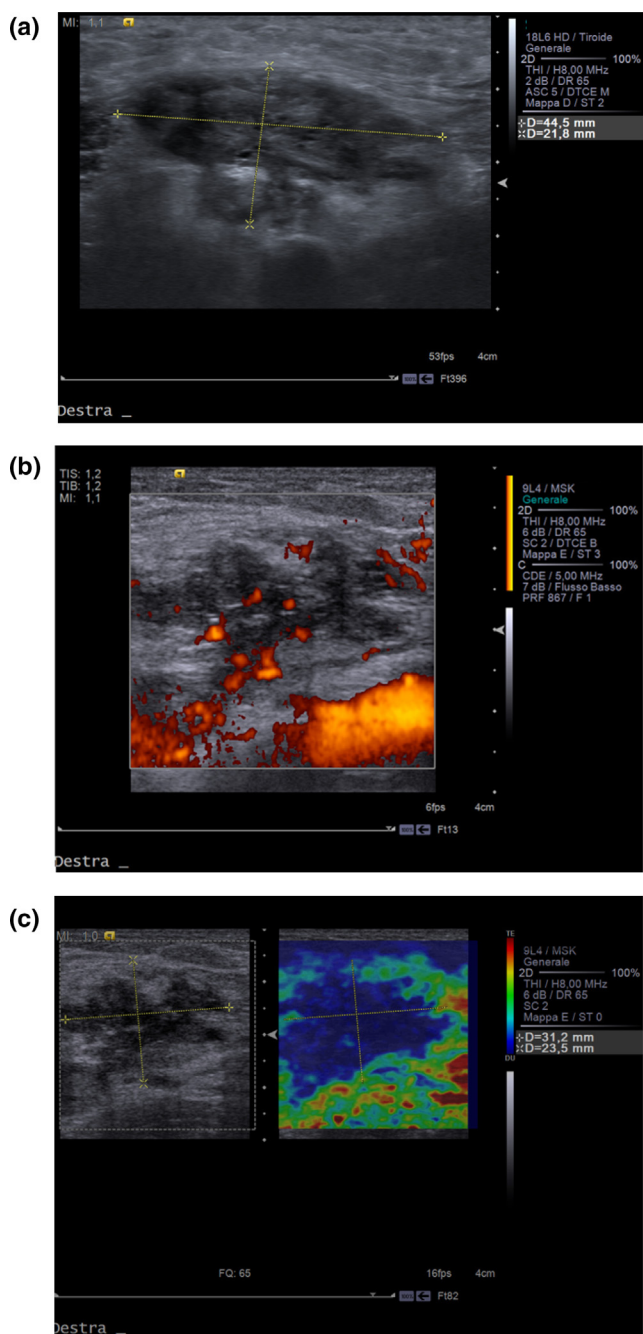
The mass, for its dimension and position, obstructed the flow of blood circulation predisposing to DVT. Indeed, despite prompt treatment with anticoagulant therapy (enoxaparin 4000 IU once daily) and elastic stocking, after 3 days the clinical condition evolved into DVT of iliac vein (Fig. 2b). Therefore, a transcatheter thrombolysis to prevent post-thrombotic syndrome was performed and complete recanalization of the vessel was obtained in 24 h. Surgical resection of the mass was performed. The patient refused hormonal therapy, to preserve her reproductive capacity.

At 6-month follow-up the patient was pain free and showed substantial improvement in her ambulation and quality of life.

## Discussion

An extensive electronic search was performed in PubMed, Scopus, Web of Science and Google. Studies were considered eligible if they described the presence of endometriosis in the context of striated muscles.

Extrapelvic endometriosis is an uncommon condition but can involve nearly every organ, resulting in a wide range of clinical manifestations. The symptoms are often nonspecific and do not correlate with extent or severity of the disease [1–5]. The presented patient had a very rare localization of endometriosis and an uncommon complication, the



**Fig. 1** (a) Solid fusiform hypoechoic mass with an inhomogeneous echotexture and irregular borders (diameter  $4.4 \times 2.18$  cm). Localization: in the right inguinal region, adjacent to subcutaneous fat, beneath the fascia of external oblique muscle, extended to iliac vein. (b) Doppler ultrasound: diffuse periphery vascularization. (c) Shear wave elastographic evaluation: Higher elasticity of the peripheral regions of the lesion (yellow-green). Homogeneous blue color distribution (hard stiffness) of the central part of lesion.

thrombosis of iliac vessel. This rare clinical and radiological presentation of extrapelvic endometriosis has not been yet reported in the literature and made diagnosis difficult.

Moreover, US can provide an evaluation of tissue stiffness through elastography. The measurement of tissue elasticity has been reported to be useful for the diagnosis and differentiation of tumors, which are stiffer than normal tissues.

Many studies described endometriosis involving the abdominal wall: within the rectus abdominis muscle and the surgical scar of previous cesarean incision [1,6–10]. The involvement of other striated muscles was rarer. Literature review reported only six cases of striated muscle affected by endometriosis: transversus abdominis [6], adductor muscles compartment [11], soleus and gastrocnemius muscles [12], piriform muscle [13], trapezius muscle [14], and deltoid muscle [15]. This paper described a case of endometriosis not previously described. The disease was localized in the subfascial layer of external oblique muscle and was the cause of an unexpected complication, due to its particular localization: a DVT of iliac vein.

The relationship between endometriosis localization and complications is not always easy to understand. A literature review of extraperitoneal endometriosis revealed varying presentations of the disease [1–15]. Nevertheless, only a very limited number of cases showed involvement of iliac vessels [3–5] and only two of them had concurrent vascular complication [4,5]. The sudden onset of symptomatology without previous associated malaise, the extension and the associated complications of the reported case was different. In 1977, the Authors Recalde and Majmudar [3] reported the first case of endometriosis involving the adventitial layer of the left femoral vein. The patient presented an irreducible groin mass, that ached and increased its size with each menstrual cycle. The case of Rosengarten et al. [4] was the first description of DVT due to endometriosis encircling the right external iliac vein. In the case described by Sharma et al. [5], the disease had a retroperitoneal localization and DVT of the right common femoral vein, external iliac and distal internal iliac veins occurred. These three patients had no previous history of a DVT and presented the typical cyclic symptomatology of endometriosis, but no previous general symptoms such as episodes of abdominal and pelvic pain, menorrhagia, metrorrhagia, dyspareunia, and dysmenorrhea. They complained of progressive swelling of the leg and limitation of function concomitant with menstruation. Instead, the *described* patient developed a sudden symptomatology, different from the cyclic catamenial edema of the other patients. In the case described by Sharma et al. [5] the patient did not take any anticoagulation therapy and developed DVT. Notwithstanding Rosengarten et al. [4] reported the development of DVT despite administration of appropriate medical therapy, similar to what was observed in this case.

The menstrual timing of the symptoms, that may have suggested the diagnosis of endometriosis, was absent in this case. So, a case similar has not been reported previously in the literature.

Since endometriotic lesions could present as a mass lesion, it seemed feasible to investigate them by the noninvasive method of fine-needle aspiration cytology (FNAC). Furthermore, ultrasound-guided biopsy, proved to be effective, fast and safe.

## Conclusions

Extrapelvic endometriosis is an uncommon condition but can involve nearly every organ, resulting in a wide range of clinical manifestations. When the localization is less common, the diagnosis is more difficult and the abdominal US can be fundamental.



**Fig. 2** (a) Mass of 22 mm irregular thickening, with spiculated margins, located in the subfascial of external oblique muscle. (b) DVT of iliac vein from extrinsic compression of the right vessels.

An uncommon localization of the disease, was observed in the external oblique muscle of patient described in this paper. Moreover, the patient developed a DVT of iliac vessel, which is a very rare complication associated with endometriosis. Likewise, the presentation of the condition was atypical with a sudden onset and without cyclical symptoms, that is unlikely for endometriosis. A similar case has not been previously reported. An extensive literature review of endometriosis involving striated muscles, was conducted addressing special attention to associated vascular complications. Finally, US had an essential role in detecting and localizing endometriosis, and providing in definitive histological diagnosis through ultrasound-guided biopsy.

#### Conflict of Interest

*The authors declare that they have no competing interests.*

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#### References

- [1] Mostafa HA, Saad JH, Nadeem Z, Alharbi F. Rectus abdominis endometriosis. A descriptive analysis of 10 cases concerning this rare occurrence. *Saudi Med J* 2013;34(10):1035–42.
- [2] A Hodnett P, McSweeney SE, Bogue C, Kelly D, Redmond HP, Maher MM. Subcutaneous endometrial deposit: an unusual cause of right iliac fossa pain. *Br J Hosp Med (Lond)* 2009;70(3):170–1.
- [3] Recalde AL, Majmudar B. Endometriosis involving the femoral vein. *South Med J* 1977;70:69–74.
- [4] Rosengarten AM, Wong J, Gibbons S. Endometriosis causing cyclic compression of the right external iliac vein with cyclic edema of the right leg and thigh. *J Obstet Gynaecol Can* 2002;24(1):33–5.
- [5] Sharma RP, Delly F, Marin H, Sturza S. Endometriosis causing lower extremity deep vein thrombosis – case report and review of the literature. *Int J Angiol* 2009;18(4):199–202.
- [6] Picod G, Boulanger L, Bounoua F, Leduc F, Duval G. Abdominal wall endometriosis after caesarean section: report of fifteen cases. *Gynecol Obstet Fertil* 2006;34(1):8–13.
- [7] Toullalan O, Baqué P, Benchimol D, Bernard JL, Rahili A, Gillet JY, et al. Endometriosis of the rectus abdominis muscles. *Ann Chir* 2000;125(9):880–3.

- [8] Wiesner W, Knespova L, Hauser M. What is your diagnosis? Extragenital endometriosis in the left rectus abdominis muscle. *Praxis* 2000;89(4):121–3 (Bern 1994).
- [9] Crespo R, Puig F, Marquina I. Pyramidalis muscle endometriosis in absence of previous surgery. *Int J Gynaecol Obstet* 2005;89(2):148–9.
- [10] Granese R, Cucinella G, Barresi V, Navarra G, Candiani M, Triolo O. Isolated endometriosis on the rectus abdominis muscle in women without a history of abdominal surgery: a rare and intriguing finding. *J Minim Invasive Gynecol* 2009;16(6):798–801.
- [11] Fambrini M, Andersson KL, Campanacci DA, Vanzi E, Bruni V, Buccoliero AM, et al. Large-muscle endometriosis involving the adductor tight compartment: case report. *J Minim Invasive Gynecol* 2010;17(2):258–61.
- [12] Poli-Neto OB. Endometriosis of the soleus and gastrocnemius muscles. *Fertil Steril* 2009;91(4):1294.
- [13] Hickey NA, Murphy JP, Bloom C, Hamilton P. Magnetic resonance imaging of endometriosis of the piriform muscle causing sciatica: case report. *Can Assoc Radiol J* 1999;50(1):33–6.
- [14] Gennari L, Luciani L. A case of endometriosis of the trapezius muscle. *Tumori* 1965;51(5):361–5.
- [15] Aron Se. Endometriosis in the region of the deltoid muscle. *Arkh Patol* 1957;19(7):67–8.