Ovarian vein thrombosis in the nonpregnant woman: an overlooked diagnosis

Kassem Harris, Suchita Mehta, Edward Iskhakov, Michel Chalhoub, Theodore Maniatis, Frank Forte and Homam Alkaied

Ther Adv Hematol
(2012) 3(5) 325–328
DOI: 10.1177/2040620712450887

Correspondence to:
Homam Alkaied, MD
Staten Island University Hospital, 475 Seaview Avenue, Staten Island, NY 10305, USA
homam.md11@yahoo.com

Kassem Harris, MD, Suchita Mehta, MD, Edward Iskhakov, MD, Michel Chalhoub, MD, Theodore Maniatis, MD and Frank Forte, MD
Staten Island University Hospital, Staten Island, NY, USA

Ovarian vein thrombosis (OVT) is a rare condition occurring in 1/600 to 1/2000 pregnancies [Dunnihoo *et al.* 1991; Ortin *et al.* 2005] mainly in the postpartum setting. It is also known to be associated with other conditions such as malignancy, pelvic inflammatory disease, inflammatory bowel disease, sepsis and recent pelvic or abdominal surgery [Andre *et al.* 2004; Heavrin and Wrenn, 2008; Jacoby *et al.* 1990; Klima and Snyder, 2008; Marcovici and Goldberg, 2000; Salomon *et al.* 1999; Simons *et al.* 1993]. It is extremely rare to find OVT without identified etiology and, hence, idiopathic OVT is only described as case reports throughout the literature. Here, we report a unique case of idiopathic isolated OVT that presented with right flank pain and an abdominal mass. Although four similar cases of idiopathic isolated OVT have been reported in the literature [Heavrin and Wrenn, 2008; Murphy and Parsa, 2006; Stafford *et al.* 2010; Yildirim *et al.* 2005], none of these patients presented with an abdominal mass. The diagnosis of isolated OVT requires a high index of suspicion. If misdiagnosed, OVT can lead to potentially fatal complications such as sepsis and pulmonary embolism. [Benfayed *et al.* 2003; Kominiarek and Hibbard, 2006; Maldjian and Zurlow, 1997; Wysokinska *et al.* 2006].

A 53-year-old postmenopausal woman with a past medical history of hypertension presented to the medical clinic complaining of 1-week history of aching right flank pain that was not associated with fever, dysuria, hematuria, nausea, vomiting, diarrhea or vaginal discharge. The patient denied any other constitutional symptoms. She is a nonsmoker with no family history of hematologic disorders. On physical examination, she was afebrile, normotensive, without tachycardia. Pelvic exam revealed a nontender, normal size uterus and adnexa. However, a 3 cm tender mass was palpated in the right lower quadrant. Laboratory data revealed a white blood cell count of 4400-cells/mm^3 and hemoglobin level 11.9 g/dl. Renal function and electrolytes were within normal limits. Computed tomography (CT) of the abdomen and pelvis with intravenous contrast showed right ovarian vein thrombus without extension to the inferior vena cava (IVC) (Figure 1).

Further work up for hypercoagulability was negative. Age- and sex-appropriate cancer screenings were all negative. Moreover, screening for ovarian pathology, with pelvic ultrasound and CA-125, was also normal. Shortly after the diagnosis of isolated OVT, the patient was placed on oral anticoagulation. It was elected not to administer antibiotics. Warfarin was continued for 5 months with the International Normalized Ratio (INR) maintained between 2 and 3. A follow-up CT scan of the abdomen and pelvis performed 5 months later showed persistence of the thrombus with no further extension beyond the ovarian vein (see Figure 2). Anticoagulation was discontinued at this point with close clinical follow up.

Ovarian vein thrombosis was first described by Austin in 1956 [Austin, 1956]. It occurs in the right side in 70–90% of cases, and bilaterally in 11–14% [Baran and Frisch, 1987; Prieto-Nieto *et al.* 2004]. The most widely accepted hypothesis for the higher incidence on the right is that the right ovarian vein is longer than the left, and lacks competent valves. The typical presentation is the triad of pelvic pain, fever, and a right-sided abdominal mass [Dessole *et al.* 2003; Dunnihoo *et al.* 1991; Klima and Snyder, 2008; Prieto-Nieto *et al.* 2004]. Fever is present in 80% and right iliac fossa pain in 55% of the patients [Prieto-Nieto *et al.* 2004]. Given the nonspecific presenting symptoms, prompt diagnosis of OVT requires a high index of suspicion. The differential diagnosis includes most conditions that affect the abdominal lower quadrant such as acute appendicitis and inflammatory bowel diseases. Therefore, imaging

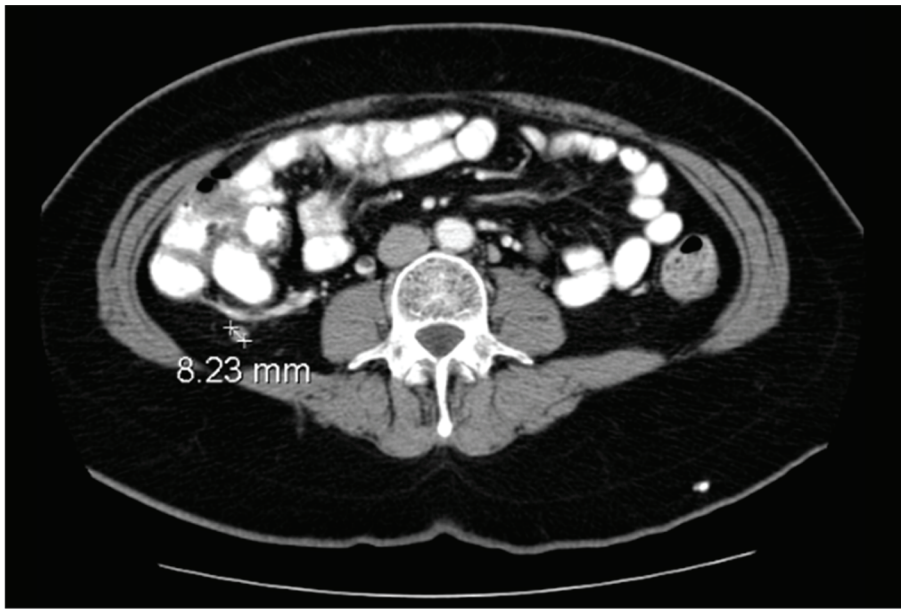

Figure 1. CT scan at presentation.

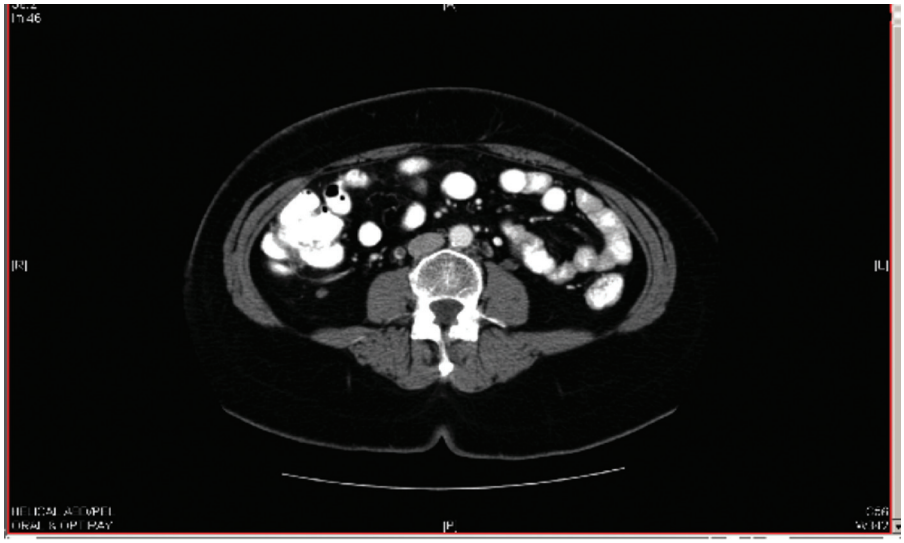

Figure 2. CT scan after 5 months of anticoagulation therapy.

studies are essential to establish the diagnosis of OVT. Magnetic resonance angiography (MRA) has the highest sensitivity and specificity that approaches 100%. CT scan with intravenous contrast enhancement has a sensitivity of 77.8% and specificity of 62.5%. Color Doppler ultrasound has the lowest sensitivity of 55.6% and a specificity of 41.5% among other imaging modalities [Kubik-Huch *et al.* 1999].

A delay in the diagnosis and treatment of OVT can lead to potentially life-threatening complications, such as thrombus extension into the IVC or ileofemoral vessels and eventually the evolution of pulmonary arterial embolization. The incidence of pulmonary embolism is approximately 25% in patients with untreated OVT and the mortality in these patients can reach about 4% [Benfayed *et al.* 2003; Dunnihoo *et al.* 1991; Kominiarek and Hibbard, 2006]. Other serious complications include septic thrombophlebitis and, rarely, infectious emboli [Dessole *et al.* 2003; Heavrin and Wrenn, 2008]. Ovarian vein thrombosis can resolve spontaneously but considering the potential catastrophic consequences, anticoagulation is usually recommended [Wysokinska *et al.* 2006].

There is no definite guideline regarding the duration of anticoagulation therapy. Wysokinska and colleagues studied the incidence and the recurrence of OVT compared with lower extremity deep venous thrombosis (DVT) [Wysokinska *et al.* 2006]. None of the 35 patients in the OVT group was idiopathic and the recurrence rate was comparable to patients diagnosed with lower extremity DVT (3 per 100 patient years of follow up). The average treatment with warfarin was 5.3 and 6.9 months for OVT and lower extremity DVT, respectively. Based on these findings, the authors suggested the application of lower extremity guidelines for the treatment of OVT.

Antibiotics can also be administered for approximately 7 days especially in cases of postpartum OVT [Brown and Munsick, 1971; Dessole *et al.* 2003; Maldjian and Zurlow, 1997; Wysokinska *et al.* 2006]. In patients with hypercoagulable disorders, anticoagulation may need to be lifelong therapy [Wysokinska *et al.* 2006]. In rare cases of persistent OVTs, an IVC filter or surgical intervention to ligate the ovarian vein can be considered [Carr and Tefera, 2006; Clarke and Harlin, 1999].

Our patient was diagnosed with an idiopathic OVT since none of the above predisposing factors for OVT were found. The patient's abdominal pain subsided few days after starting anticoagulation and she did not develop any worrisome signs such as fever, dyspnea, or chest pain. Five years later, the patient remains asymptomatic while off anticoagulation, without any further thrombotic conditions.

To date, four cases of idiopathic OVT were described [Heavrin and Wrenn, 2008; Murphy and Parsa, 2006; Stafford *et al.* 2010; Yildirim *et al.* 2005]. None of these cases had abdominal or pelvic palpable masses at presentation. Therefore, our report describes a unique case of idiopathic OVT presenting with one symptom and one sign of the typical triad. The palpable mass in the right iliac fossa was only described in cases of OVT that occur in the postpartum period as well as in other inflammatory and hypercoagulable conditions.

OVT is a rare condition with potential life-threatening complications. In female patients presenting with lower quadrant pain, with or without fever or palpable abdominal or pelvic mass, OVT should be considered in the differential diagnosis after ruling out other common conditions. MRA and CT scan with intravenous contrast are the most useful imaging modalities to diagnose this condition. Overlooking this diagnosis can lead to life-threatening conditions, such as pulmonary embolism, sepsis, and even death. Hence, prompt diagnosis of OVT requires a high index of suspicion in order to prevent these outcomes.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Conflict of interest statement

The authors declare no conflicts of interest in preparing this article.

References

Andre, M., Delevaux, I., Amoura, Z., Corbi, P., Courthaliac, C., Aumaitre, O. *et al.* (2004) Ovarian vein thrombosis in the antiphospholipid syndrome. *Arthritis Rheum* 50: 183–186.

Austin, O.G. (1956) Massive thrombophlebitis of the ovarian veins; a case report. *Am J Obstet Gynecol* 72: 428–429.

Baran, G.W. and Frisch, K.M. (1987) Duplex Doppler evaluation of puerperal ovarian vein thrombosis. *AJR Am J Roentgenol* 149: 321–322.

Benfayed, W.H., Torreggiani, W.C. and Hamilton, S. (2003) Detection of pulmonary emboli resulting from ovarian vein thrombosis. *AJR Am J Roentgenol* 181: 1430–1431.

Brown, T.K. and Munsick, R.A. (1971) Puerperal ovarian vein thrombophlebitis: a syndrome. *Am J Obstet Gynecol* 109: 263–273.

Carr, S. and Tefera, G. (2006) Surgical treatment of ovarian vein thrombosis. *Vasc Endovascular Surg* 40: 505–508.

Clarke, C.S. and Harlin, S.A. (1999) Puerperal ovarian vein thrombosis with extension into the inferior vena cava. *Am Surg* 65: 147–150.

Dessole, S., Capobianco, G., Arru, A., Demurtas, P. and Ambrosini, G. (2003) Postpartum ovarian vein thrombosis: an unpredictable event: two case reports and review of the literature. *Arch Gynecol Obstet* 267: 242–246.

Dunnihoo, D.R., Gallaspy, J.W., Wise, R.B. and Otterson, W.N. (1991) Postpartum ovarian vein thrombophlebitis: a review. *Obstet Gynecol Surv* 46: 415–427.

Heavrin, B.S. and Wrenn, K. (2008) Ovarian vein thrombosis: a rare cause of abdominal pain outside the peripartum period. *J Emerg Med* 34: 67–69.

Jacoby, W.T., Cohan, R.H., Baker, M.E., Leder, R.A., Nadel, S.N. and Dunnick, N.R. (1990) Ovarian vein thrombosis in oncology patients: CT detection and clinical significance. *AJR Am J Roentgenol* 155: 291–294.

Klima, D.A. and Snyder, T.E. (2008) Postpartum ovarian vein thrombosis. *Obstet Gynecol* 111: 431–435.

Kominiarek, M.A. and Hibbard, J.U. (2006) Postpartum ovarian vein thrombosis: an update. *Obstet Gynecol Surv* 61: 337–342.

Kubik-Huch, R.A., Hebisch, G., Huch, R., Hilfiker, P., Debatin, J.F. and Krestin, G.P. (1999) Role of duplex color Doppler ultrasound, computed tomography, and MR angiography in the diagnosis of septic puerperal ovarian vein thrombosis. *Abdom Imaging* 24: 85–91.

Maldjian, P.D. and Zurlow, J. (1997) Ovarian vein thrombosis associated with a tubo-ovarian abscess. *Arch Gynecol Obstet* 261: 55–58.

Marcovici, I. and Goldberg, E. (2000) Ovarian vein thrombosis associated with Crohn's disease: a case report. *Am J Obstet Gynecol* 182: 743–744.

Murphy, C.S. and Parsa, T. (2006) Idiopathic ovarian vein thrombosis: a rare cause of abdominal pain. *Am J Emerg Med* 24: 636–637.

Ortin, X., Ugarriza, A., Espax, R.M., Boixadera, J., Llorente, A., Escoda, L. *et al.* (2005) Postpartum ovarian vein thrombosis. *Thromb Haemost* 93: 1004–1005.

Prieto-Nieto, M.I., Perez-Robledo, J.P., Rodriguez-Montes, J.A. and Garci-Sancho-Martin, L. (2004) Acute appendicitis-like symptoms as initial presentation of ovarian vein thrombosis. *Ann Vasc Surg* 18: 481–483.

Salomon, O., Apter, S., Shaham, D., Hiller, N., Bar-Ziv, J., Itzchak, Y. *et al.* (1999) Risk factors associated with postpartum ovarian vein thrombosis. *Thromb Haemost* 82: 1015–1019.

Simons, G.R., Piwnica-Worms, D.R. and Goldhaber, S.Z. (1993) Ovarian vein thrombosis. *Am Heart J* 126: 641–647.

Stafford, M., Fleming, T. and Khalil, A. (2010) Idiopathic ovarian vein thrombosis: a rare cause of pelvic pain - case report and review of literature. *Aust N Z J Obstet Gynaecol* 50: 299–301.

Wysokinska, E.M., Hodge, D. and McBane, R.D., II (2006) Ovarian vein thrombosis: incidence of recurrent venous thromboembolism and survival. *Thromb Haemost* 96: 126–131.

Yildirim, E., Kanbay, M., Ozbek, O., Coskun, M. and Boyacioglu, S. (2005) Isolated idiopathic ovarian vein thrombosis: a rare case. *Int Urogynecol J Pelvic Floor Dysfunct* 16: 308–310.