

Pulmonary Embolism and Uterine Venous Plexus Thrombosis in the Postpartum Period

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Pulmonary thromboembolism (PTE) associated with uterine venous plexus thrombosis is very rare. This was recently observed in a puerperal woman without significant medical history. The woman had two pregnancy-associated risk factors for venous thromboembolism, which were obesity and cesarean delivery. A day after cesarean delivery, she presented with dyspnea and cyanosis. When transferred to a larger hospital, no pulse was detected, and she was pronounced dead. Autopsy examination revealed that the cause of death was related to PTE, apparently due to thrombi that originated in the uterine venous plexus.

Key Words: Pulmonary embolism; Venous thrombosis; Pregnancy; Autopsy

Introduction

Pregnancy is associated with an increased risk of venous thromboembolism, which includes pulmonary embolism (PE) and deep vein thrombosis. The incidence of venous thromboembolism is estimated at 0.76 to 1.72 per 1,000 pregnancies, which is four times higher than that in non-pregnant women [1]. The incidence of PE in the postpartum period is much higher

than during pregnancy [2]. There are few reports on uterine vein thrombosis, consisting mostly of cases involving thrombi in external and internal iliac veins [3]. Uterine venous plexus thrombosis (UVPT) is a rare diagnosis. Sudden death linked to pulmonary thromboembolism (PTE) in the presence of UVPT without deep vein thrombosis in the lower extremities is even more uncommon. We report a case of sudden, unexpected death due to acute PTE associated with UVPT in a postpartum woman.

Case Report

A 34-year-old woman with a prior history of cesarean delivery was admitted to a local hospital at 38 weeks of gestation. Her previous pregnancy was uncomplicated, and she had no significant medical history. A day after admission, she underwent cesarean delivery. The newborn baby weighed 3,520 g at birth, with an Apgar score of 9/10. During surgery, no abnormal symptoms were observed. Twenty-seven hours after the surgery, the woman presented with acute-onset dyspnea and dizziness. Her vital signs indicated a slightly elevated blood pressure (150/100 mm Hg), tachypnea (respiratory rate, 27 breaths per minute), tachycardia (heart rate, 126 beats per minute), and normal temperature (36.8°C). She was transferred to a nearby larger hospital. Cyanosis was seen on her face, hands, and feet. No pulse was detected and cardiopulmonary resuscitation was consequently performed. Two hours after the procedure, she died due to sudden cardiopulmonary arrest.

The patient autopsied was an extremely obese woman 162 cm in height and 95 kg in weight (body mass index, 36.2). External examination showed no evidence of injury, except bruises in the anterior chest

(due to cardiopulmonary resuscitation). Internal examination revealed a horizontal fracture in the sternum and multiple bilateral anterior fractures from the second to the fifth ribs (due to cardiopulmonary resuscitation). The right and left lung weighed 468 g and 642 g, respectively. The cut surface of the right lung revealed marked bloody congestion and edema with a thrombus filling the right pulmonary artery (Fig. 1). No findings were identified in the left lung or associated vessels. The uterus weighed 1,394 g. One intramural mass, measuring 3.5 × 3.0 cm in size, was identified. The right uterine venous plexus revealed a thrombus, measuring 1.5 × 1.0 cm in size (Fig. 2). Findings of the remaining internal organs were unremarkable. There was no evidence of deep vein thrombosis in the lower extremities. Microscopic examination confirmed a thrombus that filled the right pulmonary artery, in addition to congestion and edema in the right lung. The combined evidence suggests that the cause of death was PTE, possibly associated with UVPT.

Discussion

The literature supports an association between deep vein thrombosis of the lower limbs and PE. During pregnancy, the uterus increases in size and has

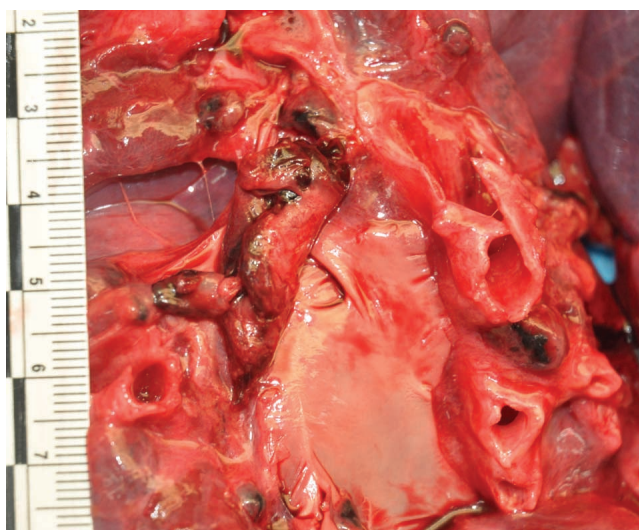


Fig. 1. A large thrombus was visible in the right pulmonary artery.



Fig. 2. A thrombus was identified in the uterine venous plexus.

increased blood flow [3]. These changes may impede venous outflow from the lower limbs until approximately six weeks after delivery. Pregnancy is also classically thought to be a hypercoagulable state [1]. This predisposes women to developing venous thromboembolism. Additional risk factors for venous thromboembolism include heart disease, diabetes, lupus, smoking, multiple pregnancy, age greater than 35 years, obesity, and cesarean delivery. Because of their high prevalence, age greater than 35 years, obesity, and cesarean delivery are the most important factors that can increase the incidence rates of venous thromboembolism [4,5]. A clear relationship has been established between deep vein thrombosis and PE. Deep vein thrombosis associated with PE usually occurs in deep vessels of lower limbs, especially in those of the left leg (approximately 70% to 90% of cases). Occasionally, a large uterine leiomyoma can cause compression of the pelvic veins, leading to persistent pressure on these veins, and inducing chronic venous stasis in the pelvis and lower extremities [6]. The uterine venous plexus extends along the lateral aspect of the uterus and communicates with the ovarian and vaginal plexuses within the broad ligament. They are drained by the uterine veins arising from the lower parts of the uterine plexuses, lateral to the cervix. More common forms of pelvic venous thrombosis are uterine vein thrombosis, ovarian vein thrombosis, and cervical varices [7–10]. Because an anastomosis between the ovarian and uterine venous plexuses may obscure the venous origin, a radiologic diagnosis of UVPT may be difficult [7].

The present case had no other origins of PE except UVPT. The thrombus was found within the venous plexus located in the lateral side of the lower uterus. The femoral vessels of both legs and deep vessels of both lower legs had no thrombi. Although an intramural leiomyoma was observed, it was too small to compress one of the pelvic vessels. Although PE resulting from UVPT is very rare, this patient had two risk factors for venous thromboembolism, obesity and cesarean delivery. These factors may have contributed

to the development of UVPT, finally leading to PE.

The diagnosis of proximal thrombosis involving the pelvic veins is usually made only after the thrombus extends into the veins of the lower legs [3]. UVPT may therefore be a precursor to deep vein thrombosis of the lower limbs, leading to the development of PTE. On the other hand, it must be taken into account that deep vein thrombosis of the lower limbs may extend to proximal areas and be detected as UVPT. In both cases, it can be concluded that UVPT contributes to PTE.

PTE can occur exclusively without deep vein thrombosis. For this reason, PTE and UVPT may be considered independent events that do not affect each other. However, since deep vein thrombosis is an established cause of PTE, the connection between UVPT and PTE cannot be excluded in a case of confirmed UVPT, even though UVPT is rare in itself and its association with PTE is uncommon. For this reason, we infer that PE and UVPT are related in this case.

In conclusion, PE concurrent with UVPT during pregnancy is very rare. To our knowledge, this case is the first in the literature to report sudden, unexpected death caused by acute PTE related to UVPT in a postpartum woman without deep vein thrombosis of the lower limbs. This case study suggests that UVPT can lead to PTE, and that careful examination of vessels surrounding the uterus may help discern the origin of PE in women whose lower limbs show no signs of deep vein thrombosis.

Conflicts of Interest

No potential conflict of interest relevant to this article was reported.

References

1. Marik PE, Plante LA. Venous thromboembolic disease and pregnancy. *N Engl J Med* 2008;359:2025-33.
2. Heit JA, Kobbervig CE, James AH, et al. Trends in the incidence of venous thromboembolism during pregnancy or postpartum: a 30-year population-based study. *Ann Intern Med* 2005;143:697-706.

3. Mavrellos D, Cohen H, Pateman K, et al. Diagnosis of uterine vein thrombosis on transvaginal ultrasound. *Ultrasound Obstet Gynecol* 2013;42:480-3.
4. James AH, Jamison MG, Brancazio LR, et al. Venous thromboembolism during pregnancy and the postpartum period: incidence, risk factors, and mortality. *Am J Obstet Gynecol* 2006;194:1311-5.
5. Larsen TB, Sorensen HT, Gislum M, et al. Maternal smoking, obesity, and risk of venous thromboembolism during pregnancy and the puerperium: a population-based nested case-control study. *Thromb Res* 2007;120:505-9.
6. Fernandes FL, Dinardo CL, Terra-Filho M. Uterine myoma as a cause of iliac vein thrombosis and pulmonary embolism: common disease, rare complication. *Respirol Case Rep* 2014;2:132-4.
7. Leibovitz Z, Degani S, Shapiro I, et al. Diagnosis of pregnancy-associated uterine venous plexus thrombosis on the basis of transvaginal sonography. *J Ultrasound Med* 2003;22:287-93.
8. Al-toma A, Heggelman BG, Kramer MH. Postpartum ovarian vein thrombosis: report of a case and review of literature. *Neth J Med* 2003;61:334-6.
9. Royo P, Alonso-Burgos A, Garcia-Manero M, et al. Postpartum ovarian vein thrombosis after cesarean delivery: a case report. *J Med Case Rep* 2008;2:105.
10. Sammour RN, Gonen R, Ohel G, et al. Cervical varices complicated by thrombosis in pregnancy. *Ultrasound Obstet Gynecol* 2011;37:614-6.