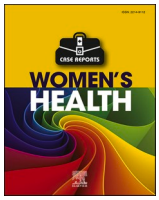
Case Reports in Women's Health 42 (2024) e00608

Contents lists available at ScienceDirect

Case Reports in Women's Health

journal homepage: www.elsevier.com/locate/crwh



Spontaneous uterine rupture complicated by bilateral pulmonary emboli: A case report

Sara Wetzler *, Camila Cabrera, Peter S. Bernstein

Department of Obstetrics, Gynecology and Reproductive Sciences, Icahn School of Medicine at Mount Sinai Hospital, New York, NY 10029, USA

ARTICLE INFO

Keywords:
Uterine rupture
Pulmonary emboli
Cesarean delivery
Unscarred uterus
Case report

ABSTRACT

Spontaneous uterine rupture in unscarred uteri complicated by pulmonary emboli is a rare event with major maternal morbidity and mortality.

This is a case of a 32-year-old woman, G1P0, at term, with no pertinent past medical/surgical history, who underwent an emergency cesarean delivery for failed induction of labor complicated by uterine rupture. Post-operatively, the patient was tachycardic and hypoxic. CT arteriogram revealed massive bilateral pulmonary emboli, and she was transferred for specialist care. An emergency pulmonary embolectomy and implantation of an extracorporeal right ventricular assist device were performed. Once the patient was clinically stable, an evaluation for thrombophilias and collagen disorders was done, and was positive for a variant of unknown significance in the ELN gene (c.205G > C).

This case report highlights a potential connection between uterine ruptures, hemorrhage, and multiple, large pulmonary emboli. The authors propose a multidisciplinary discussion and evaluation to identify risk factors and biologic causes for these rare but life-threatening complications.

1. Introduction

Spontaneous uterine rupture in an unscarred uterus is a rare event that can lead to maternal and fetal morbidity and mortality. The incidence of rupture of unscarred uteri is 0.0175% to 0.005% of pregnancies [1]. Causes of uterine rupture include trauma, congenital weakness of the myometrium (e.g., Ehlers-Danlos type IV) [2], acquired weakness secondary to prolonged labor or uterotonic drugs [3,4], or uterine cavity overdistension due to multiple gestation, high parity, or uterine anomalies [1].

Massive thrombo-emboli during pregnancy and labor are also rare events, with an incidence rate of about 1 in 1000–3000 pregnancies [5]. The hypercoagulable state of pregnancy with increased levels of prothrombotic factors increases the risk for venous thromboembolism (VTE) [5]. Other risk factors include previous thrombosis, thrombophilia, rheumatoid disease, inflammatory bowel disease, obesity, immobilization, preeclampsia, (emergency) cesarean delivery (CD), and major postpartum bleeding [6,7].

Patients with uterine rupture often suffer from hemorrhage and require large blood transfusions. Studies have demonstrated increased thromboembolic risk associated with transfusions [8,9]. However, a large retrospective cohort study found the addition of fresh frozen plasma was more protective than transfusions of red blood cell alone [10].

While there are many articles reporting cases of uterine rupture in unscarred uteri [11–13] and many case reports of pulmonary embolism (PE) after a CD [14–17], there are apparently no articles describing a case of uterine rupture in an unscarred uterus followed by massive PE. Here, with consent provided by the patient, a case of uterine rupture following failed induction of labor at term complicated by massive bilateral PE is described. Management required transfer for specialist care after cesarean section.

2. Case Presentation

A 32-year-old woman, G1P0, with no pertinent past medical or surgical history, underwent an induction of labor at 40w4d for gestational hypertension at an initial hospital.

Her obstetrical history was significant for elevated 50 g glucose challenge test followed by a normal 100 g glucose tolerance test. She had had an incomplete anatomy sonogram at that hospital but reported a normal anatomy sonogram at 31 weeks. She was GBS positive. Her

* Corresponding author at: 1176 5th Ave, Klingenstein Pavilion 9th floor, New York, NY 10029, USA.
E-mail address: sara.wetzler@icahn.mssm.edu (S. Wetzler).

https://doi.org/10.1016/j.crwh.2024.e00608
Received 11 March 2024; Received in revised form 4 April 2024; Accepted 9 April 2024
Available online 10 April 2024

gynecologic history included a HPV16 positive Pap smear with no intraepithelial lesion. She denied a history of fibroids, ovarian cysts, sexually transmitted infections, with no history of D&C or other gynecological-surgical procedures on the uterus.

Per documentation from the outside hospital, the patient had received oxytocin for over 21 h. While in labor, she developed a fever to 38.3C and the fetal heart tracing was significant for fetal tachycardia of 180 bpm for over 10 h with minimal variability, concerning for chorioamnionitis. About 21 h into labor, she complained of a sharp cramp in her abdomen, starting in the epigastric area, extending to the periumbilical area, and radiating to the left upper quadrant, bilateral shoulders, and right flank. A sonogram was unremarkable. Variable decelerations were observed with no cervical change past 8 cm and a primary CD was recommended for arrest of dilation and category 2 tracing remote from delivery. After receiving spinal anesthesia, there was fetal bradycardia to 50 bpm, prompting an emergency CD. Upon entry into the peritoneal cavity, a hemoperitoneum was noted.

A live male weighing 3720 g was delivered via a low-transverse uterine incision and handed to the neonatology team for resuscitation. Apgar scores were 2 at 1 min and 8 at 5 min. The uterus was exteriorized for repair, at which point a defect in the posterior uterine wall was appreciated through the anterior lower uterine segment hysterotomy. It was described as an 8 cm vertical defect approximately 4 cm left of the midline, consistent with uterine rupture of an unknown duration. It was surmised that the rupture began at the time of her abdominal/shoulder pain 3 h prior to the delivery. The rupture site was repaired with a running locked suture of 0 Monocryl. Following the hysterotomy repair, a 3 cm defect was observed in the bladder dome, repaired by the urology team. The operation lasted 3 h and the patient received 2200 cc IV crystalloid, 2 units packed red blood cells and 2 units of fresh frozen plasma, and tranexamic acid. The quantitative blood loss was 1520 cc. The placental pathology report was unavailable.

Post-operatively the patient was tachycardic to 132 bpm and hypoxic with spO2 of 80–90%, requiring 5–6 L of supplemental O2 via nasal cannula. Based on these abnormal vital signs and after a complicated delivery, the differential diagnoses included intra-abdominal hemorrhage, pulmonary edema, PE, peripartum cardiomyopathy, sepsis, or transfusion reaction. A CT arteriogram was performed to evaluate for pulmonary edema vs. PE (there was low suspicion for the other differential diagnoses), and it demonstrated extensive bilateral PE with flattening intraventricular septum consistent with right cardiac dysfunction. On echocardiogram, her right ventricle was almost akinetic. She was started on an intravenous heparin drip and transferred to the regional perinatal center's (RPC) surgical intensive care unit for emergency thrombolysis and inferior vena cava (IVC) filter placement, which was not available at the outside hospital.

On arrival at the RPC, the patient was unable to speak in full sentences, but reported chest pain and fatigue. She was tachycardic (130–140 bpm). Transthoracic echocardiogram demonstrated severely dilated right ventricle and hypokinesis. An emergency pulmonary embolectomy and implantation of an extracorporeal right ventricular assist device (RVAD) and rigid sternal fixation was performed. The main pulmonary artery was dissected, and the entire left branch pulmonary artery and the intermediate branch to the middle and lower lobes were obliterated by acute thrombi, which were retrieved (Fig. 1). Smaller thrombi in the upper and lobe segmental arteries were also removed. Cardiopulmonary bypass was weaned, and RVAD was initiated as the patient had very little cardiac ejection without its support. An IVC filter was placed.

The patient recovered in the ICU on ampicillin, gentamycin, and clindamycin for chorioamnionitis and remained intubated, sedated, on mechanical ventilation until she was extubated and weaned off RVAD on hospital day 3. She developed pelvic abscesses (treated with piperacillin/ tazobactam), but her fever persisted and, ultimately, the abscesses were drained by the interventional radiology team.

Once the patient was clinically stable, she was evaluated for thrombophilias, and collagen disorders, like Ehler's Danlos, via genetic testing. Results of the evaluation noted that the patient was wild type for Prothrombin 620210 A and Factor V Leiden and had normal levels of Anticardiolipin IgG/ IgM, IgM total, IgG total, and Beta-2-glycoprotein I AB IgG/IgA, IgM. A lupus anticoagulant panel was positive; however, false positives can occur in patients on anticoagulant medication. She was ANA negative. Genetic testing for COL3A1 gene (involved in the production of type III collagen) was also done and the patient was negative for vascular Ehlers Danlos, but reflex testing to an Ehlers Danlos syndrome panel found a VUS (variant of uncertain significance) in the ELN gene (c.205G > C). Though Ehlers Danlos syndrome was ruled out as a potential etiology for the uterine rupture, she was recommended to follow up with Genetics in 1–2 years to re-analyze the sample and reassess the significance of the VUS as more knowledge is obtained about the various gene variants. She was counseled on the increased risk of recurrent spontaneous uterine rupture, and an etonogestrel implant was placed for contraception.

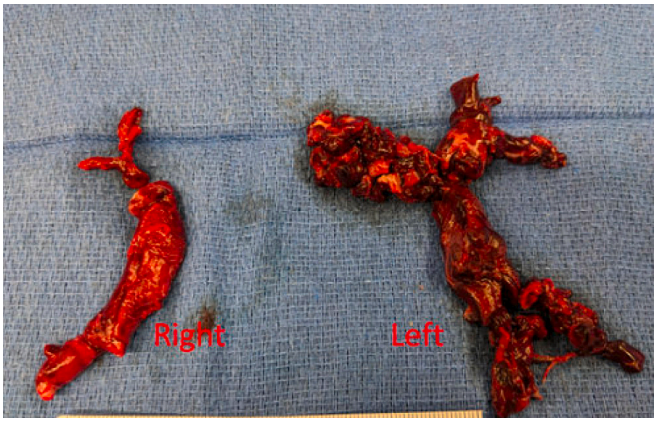


Fig. 1. Pulmonary emboli removed from the intermediate branch to the middle and lower lobes.

3. Discussion

The literature demonstrates that spontaneous uterine rupture is a rare occurrence, particularly for patients with no history of uterine surgery. What is even rarer is a combination of uterine rupture and thromboembolic events. It is unclear if these two events are associated or coincidental. Possible causes of uterine rupture in this case could be the prolonged administration of oxytocin causing acquired weakness. It could also be due to a potential unknown connective tissue disorder linked to the VUS due to the patient experiencing two organ defects (although it is unclear if the bladder defect was iatrogenic). More research into this variant is required to fully understand if it could have increased this patient's risk of organ rupture. Possible independent factors causing the massive PE following blood transfusions for hemorrhage include trauma and the emergency CD in conjunction to the hypercoagulable state secondary to pregnancy and prolonged immobility.

This case of a woman with no pertinent past medical or surgical history who experienced a uterine rupture after an induction of labor adds to the limited literature on spontaneous uterine rupture complicated by massive PE as there are no similar cases in the literature. It reflects the need to further understand causes of spontaneous uterine ruptures of unscarred uteri as well as causes of thromboemboli. The case also poses interesting questions regarding the how to work up the patient for potential etiologies, and how to counsel the patient regarding family planning following a pregnancy complication. The authors recommend multidisciplinary discussion and genetic evaluation for spontaneous uterine ruptures, identifying risk factors and biologic causes for this rare but life-threatening phenomenon. The authors also recommend careful monitoring of patients who undergo transfusion

protocols as they are a greater risk for thromboembolic events.

Contributors

Sara Wetzler contributed to patient care, conception of the case report, acquiring and interpreting the data, drafting the manuscript, undertaking the literature review, and revising the article critically for important intellectual content.

Camila Cabrera contributed to patient care, conception of the case report, interpreting the data, drafting the manuscript, and revising the article.

Peter S. Bernstein contributed to patient care, interpreting the data, drafting the manuscript, and revising the article.

All authors approved the final submitted manuscript.

Funding

No funding from an external source supported the publication of this case report.

Patient consent

Obtained.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

References

[1] K. Nanda, S. Singh, V. Dhama, R. Choudhary, R. Azad, A rare case of spontaneous posterior wall rupture in an unscarred uterus during labor, Int. J. Reprod. Contracept. Obstet. Gynecol. 6 (9) (2017) 4127–4130, https://doi.org/10.18203/2320-1770.ijrcog20174075.

[2] C. Walsh, W. Reardon, M. Foley, Unexplained pre-labor uterine rupture in a term Primigravida. [letter to the editor], Obstet. Gynecol. 109 (2) (2007) 455.

[3] I. Al-Zirqi, A.K. Daltveit, L. Forsén, B. Stray-Pedersen, S. Vangen, Risk factors for complete uterine rupture, Am. J. Obstet. Gynecol. 216 (2) (2017) 165.e1–165.e8, https://doi.org/10.1016/j.ajog.2016.10.017.

[4] J.J. Zwart, J.M. Richters, F. Ory, J.I. de Vries, K.W. Bloemenkamp, J. van Roosmalen, Uterine rupture in the Netherlands: a nationwide population-based cohort study, BJOG 116 (8) (2009) 1069–1080, https://doi.org/10.1111/j.1471-0528.2009.02136.x.

[5] G. Cueto-Robledo, F.D. Cervantes-Naranjo, L.M. Gonzalez-Hermosillo, E. Roldan-Valadez, L.E. Graniel-Palafox, K.Y. Castro-Escalante, B. Orozco-Zuñiga, Pulmonary embolism during pregnancy: an updated review with case series description, Curr. Probl. Cardiol. 48 (7) (2023) 101683, https://doi.org/10.1016/j.cpcardiol.2023.101683.

[6] R.A. Virkus, E. Løkkegaard, Ø. Lidegaard, J. Langhoff-Roos, A.K. Nielsen, K. J. Rothman, T. Bergholt, Risk factors for venous thromboembolism in 1.3 million pregnancies: a Nationwide prospective cohort, PLoS One 9 (5) (2014) e96495, https://doi.org/10.1371/journal.pone.0096495.

[7] A.H. James, Venous thromboembolism in pregnancy, Arterioscler. Thromb. Vasc. Biol. 29 (3) (2009) 326–331, https://doi.org/10.1161/ATVBAHA.109.184127.

[8] E. Grandone, D. Colaizzo, M. Mastroianno, F. Petruzzelli, L. di Mauro, M. Carella, G.L. Tiscia, A. Ostuni, Pulmonary embolism associated with transfusion after severe postpartum Haemorrhage: is less more? Blood Transfus. 18 (1) (2020) 13–19.

[9] A. Zander, E. Olson, J.-M. Van Gent, J. Bandle, R. Calvo, S. Shackford, Kimberly Peck, C. Sise, M. Sise, Bryan King, Does resuscitation with plasma increase the risk of venous thromboembolism? J. Trauma Acute Care Surg. 78 (1) (2015) 39–44.

[10] L. Thurn, A. Wikman, P. Lindqvist, Postpartum blood transfusion and hemorrhage as independent risk factors for venous thromboembolism, Thromb. Res. 165 (2018) 54–60, https://doi.org/10.1016/j.thromres.2018.03.002.

[11] S. Uccella, A. Cromi, G. Bogani, E. Zaffaroni, F. Ghezzi, Spontaneous prelabor uterine rupture in a primigravida: a case report and review of the literature, Am. J. Obstet. Gynecol. 205 (5) (2011) e6–e8, https://doi.org/10.1016/j.ajog.2011.08.013.

[12] S.D. Halassy, J. Eastwood, J. Prezzato, Uterine rupture in a gravid, unscarred uterus: a case report, Case Rep. Women's Health 24 (2019) e00154, https://doi.org/10.1016/j.crwh.2019.e00154.

[13] V. Revicky, A. Muralidhar, S. Mukhopadhyay, T. Mahmood, A case series of uterine rupture: lessons to be learned for future clinical practice, J. Obstet. Gynaecol. India 62 (6) (2012) 665–673, https://doi.org/10.1007/s13224-012-0328-4.

[14] B. Song, Y. Sun, D. Liu, G. Li, Acute pulmonary embolism immediately after cesarean section despite dilatation of the left ventricle: a case report and literature review, Emerg. Crit. Care Med. 3 (3) (2023) 130–135, https://doi.org/10.1097/EC9.0000000000000073.

[15] K.H. Ahn, S.C. Hong, Embolectomy for massive pulmonary embolism after cesarean delivery, CMAJ 188 (4) (2016) E73, https://doi.org/10.1503/cmaj.150176.

[16] A.D.B. Vuong, T.H. Pham, V.H. Bui, X.T. Nguyen, N.B. Trinh, Y.O.N. Nguyen, D.K. T. Le, P.N. Nguyen, Successfully conservative management of the uterus in acute pulmonary embolism during cesarean section for placenta previa: a case report from Tu Du Hospital, Vietnam and literature review, Int. J. Emerg. Med. 17 (14) (2024), https://doi.org/10.1186/s12245-024-00587-4.

[17] J. Park, S.C. Hong, H.Y. Yun, Y. Jeon, S. Kim, Song, & S.W., Massive pulmonary embolism after cesarean section managed with surgical thrombectomy bridged with extracorporeal membrane oxygenation: a case report, Int. J. Surg. Case Rep. (2023) 107, https://doi.org/10.1016/j.ijscr.2023.108371.