

Transvaginal Ultrasound-guided Thrombin Injection for the Treatment of Secondary Postpartum Hemorrhage Caused by a Pseudoaneurysm of the Uterine Artery

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Introduction

Postpartum hemorrhage is the most important cause of maternal death worldwide, and the most important cause of maternal morbidity in The Netherlands [1]. Postpartum hemorrhage can be differentiated into a primary (early) hemorrhage and secondary (late) hemorrhage. Primary postpartum hemorrhage develops in the first 24 hours postpartum and is usually caused by one of 'the four T's': Tonus (uterine atony), Tissue (retained pregnancy products), Trauma (rupture), and Thrombin (coagulopathy). Secondary postpartum hemorrhage develops between 24 hours and twelve weeks postpartum. The incidence of secondary postpartum hemorrhage is 2% in developed countries [2]. The most frequent causes are retained pregnancy products, subinvolution of the uterus, coagulopathy, and endometritis. More rare causes of a secondary postpartum hemorrhage are persistent trophoblastic disease, arteriovenous malformation, and a pseudoaneurysm of the uterine artery. We describe a 33-year old patient who presented six weeks postpartum in December 2014 at the emergency department of the Albert Schweitzer Hospital in The Netherlands, with severe vaginal bleeding caused by a pseudoaneurysm of the uterine artery as a rare complication after Caesarean section. The pseudoaneurysm was initially treated by embolization of the uterine artery, unfortunately without adequate effect. Definitive treatment was obtained by transvaginal ultrasound-guided thrombin injection into the uterine artery and pseudoaneurysm.

Keywords: Pseudoaneurysm; Secondary postpartum hemorrhage; Embolization; Thrombin injection

Case Report

We describe the course of delivery and late complications of the second birth of a 33-year-old woman, with an uncomplicated obstetric medical history. She underwent a diagnostic laparoscopy because of endometriosis, dilation and curettage for a missed abortion, and a primary Caesarean

section for her first baby because of fetal breech position. All procedures and recoveries were uneventful. The course of her current pregnancy was uneventful, there was a spontaneous onset of labor at a gestational age of 37 weeks and she attempted a vaginal birth after prior Caesarean. She ultimately underwent a secondary Caesarean section because of arrest of cervical dilation. During surgery, the rectus abdominis muscle was incised because of a difficult extraction of the child. Arterial bleeding was noted at the left side of the incision, which was surgically treated by suture ligation. Sulprostone 500 µg was administered intravenously in two hours because of primary postpartum hemorrhage. Total blood loss was 1100 cc. One day postpartum her hemoglobin level was 3.9 mmol/l, and she received two units of packed red blood cells. She was discharged one day later.

Six weeks after Caesarean section she presented at our emergency clinic department with severe vaginal bleeding and hypotension (80/40 mmHg). After hemodynamic stabilization she underwent dilation and curettage. Total blood loss was 800 cc. The next day she was discharged. The tissue retained during curettage revealed no evidence of retained pregnancy products, only clots.

Nine weeks postpartum she returned to our emergency department with severe vaginal bleeding with a total blood loss estimated to be 2 liters, and a blood pressure of 70/50 mmHg. She was hemodynamically stabilized and Tranexamic acid 1000 mg was administered. On transvaginal ultrasound an extremely retroverted uterus was seen and an endometrial thickness of 15 mm. Little free fluid was seen in the rectouterine pouch. Screening for coagulation disorders turned out negative. MRI of the lower abdomen (**Figure 1**) showed at the left side of the uterine incision a subserosal cavity filled with hemorrhage, which continued into the cervical canal.

Furthermore, there were blood products intra-abdominally from the lateral aspect of the uterus up to the psoas muscle. There was also a hemorrhagic cyst of the right adnex. Under the working diagnosis of first menstruation postpartum, oral

contraceptive and antibiotics were started. Four days later she was discharged home in good clinical condition.

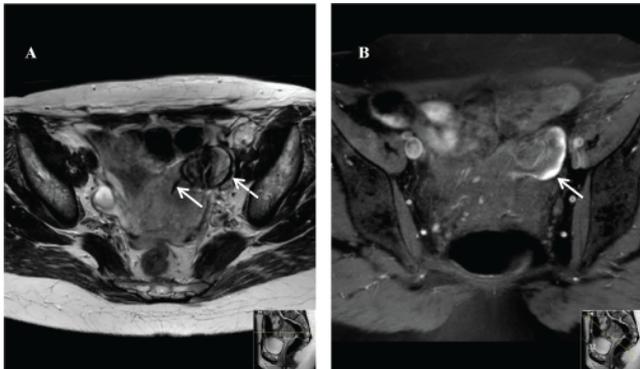


Figure 1: A) MRI (T2 WI) of the lower abdomen showing subserosal cavity with hypointense lining (dotted arrow) indicating blood products, continuing into the uterine incision (straight arrow). B) MRI (T1 WI, fat sat) of the lower abdomen showing subserosal cavity with hyperintense lining (straight arrow) indicating the presence of hemosiderine.

Our patient returned to the hospital only two days later with massive vaginal bleeding, and again Tranexamic acid 1000 mg was started. She received two units of fresh frozen plasma and four units of packed red blood cells. Hemodynamic stability was achieved inadequately, and therefore a subsequent selective angiography of the left uterine artery was performed. Angiography showed a pseudoaneurysm of the uterine artery (**Figure 2A**).

A coil embolization was successfully performed of the origin of the left uterine artery after which the pseudoaneurysm did not fill with contrast agent (**Figure 2B**). She received 5 units of packed red blood cells and two units of fresh frozen plasma. Because of hemorrhagic shock she was transferred to the Intensive Care Unit. One day after embolization her hemoglobin level was 5.7 mmol/l and she was hemodynamically stable. After a week she was discharged. Unfortunately, no imaging was performed to ensure definite treatment of the pseudoaneurysm.

One day after discharge she returned because of recurrent vaginal blood loss, and a re-embolization procedure was performed of the anterior division of the internal iliac artery with coil placement just proximal and distal to the origin of the uterine artery, which could not be selectively catheterized because of the previously placed coil, spasms and dissection of the uterine artery. However, a control CT angiography the next day showed still partial filling of the pseudoaneurysm by a collateral artery originating just proximally to the placed coils (**Figure 3**).

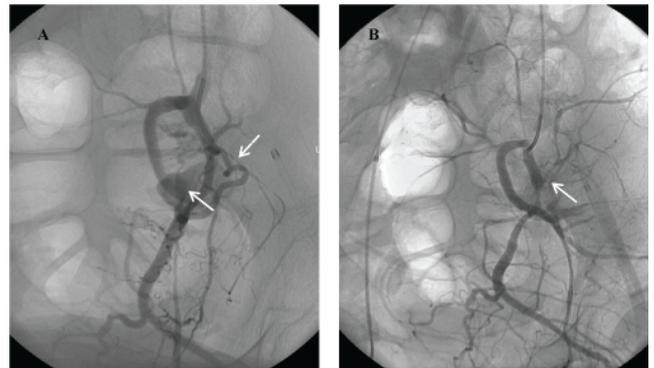


Figure 2: A) Angiography with selective contrast injection of the anterior division of the left uterine artery (dotted arrow), showing a blush representative of a pseudoaneurysm (straight arrow). B) Repeat angiography after coiling demonstrating absence of filling of the uterine artery and pseudoaneurysm with a coil at the origo of the uterine (arrow).

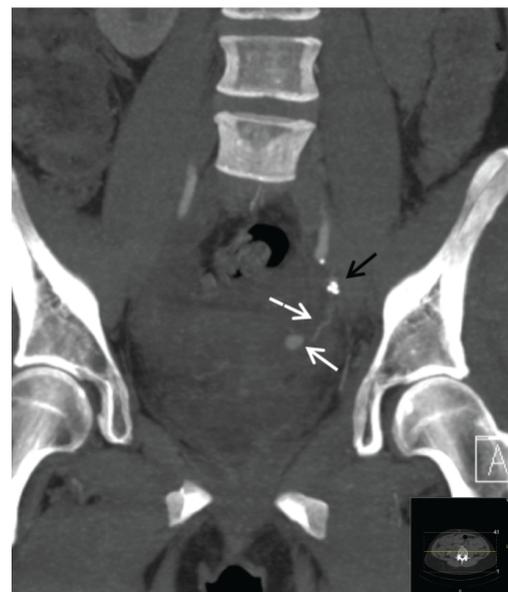


Figure 3: Computed tomography (coronal) of the lower abdomen demonstrating collateral filling (dotted arrow) of the pseudoaneurysm (straight white arrow) despite coiling at the origin of the uterine artery (black arrow).

Definitive treatment was performed by transvaginal ultrasound-guided injection of 5000 IE thrombin (D-Stat flowable hemostat, Vascular Solutions, Inc. Minneapolis, USA) into the pseudoaneurysm. Color Doppler imaging afterwards showed an occlusion of the left uterine artery and pseudoaneurysm. There has been no sign of recurrence of disease since eighteen months.

Discussion

A pseudo-aneurysm of the uterine artery is a rare and potentially fatal cause of secondary postpartum hemorrhage. Although rare, these should not be missed because of the severe consequences when missed. A thorough physical examination, noting the full patients' history and screening for coagulopathy are mandatory. Transvaginal ultrasound will provide information about possible retained pregnancy products. Absence of β -HCG will exclude a persistent trophoblastic disease. If all normal, color Doppler imaging should be performed to evaluate possible arteriovenous malformations or pseudoaneurysms. A 'yin and yang sign' or 'to-and-fro sign' at color Doppler imaging is typical for pseudoaneurysm. Angiography, CT and MRI are alternative modalities to detect a pseudoaneurysm.

An arterial pseudoaneurysm is defined by a disruption of the arterial wall where the extraluminal flow is contained by adjacent tissue of the artery. In a historical cohort reporting on 60 patients suffering from delayed postpartum hemorrhage, two patients were diagnosed with a pseudoaneurysm [3]. The formation of a pseudoaneurysm is associated with surgical trauma with vascular injury [4], but is also described after spontaneous vaginal delivery [5]. Historically hysterectomy with or without arterial ligation was the standard of care for treatment of pseudoaneurysms due to postpartum hemorrhage. Currently, the vast majority of patients are treated using selective arterial embolization [6]. Embolization is safe and has no major short- or long-term side effects [6]. Effectiveness of the procedure in this setting is estimated at 91-94% [7, 8] When adequate hemostasis cannot be achieved using embolization, hysterectomy should be considered.

Alternatively, guided by ultrasound thrombin can be injected transvaginally or percutaneously into the pseudoaneurysm (ultrasound-guided thrombin injection; UGTI). UGTI is a relatively simple procedure and has high success rates in post catheterization pseudoaneurysms, for which UGTI is the treatment of choice [9-11]. The primary success rate for this indication is 97% and the complication rate is described as approximately 1% [12]. In postpartum hemorrhage due to pseudoaneurysms the use of UGTI is not well known. The treatment was only described in two earlier case reports, both times with success [13]. We recommend that UGTI should be considered before hysterectomy in case of failure of embolization.

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