Embolization of a left uterine artery mycotic aneurysm after a neglected, perforated appendicitis before delivery

Dette Elise Anne van Doorn¹, Maarten van Leuken² & Robertus Johannes Paulus Rijnders¹

¹Department of Obstetrics and Gynaecology, Jeroen Bosch Hospital, 's-Hertogenbosch, The Netherlands
²Department of Radiology – Intervention Radiology, Jeroen Bosch Hospital, 's-Hertogenbosch, The Netherlands

Key Clinical Message
We report a case of a successful embolization of a mycotic aneurysm of the left uterine artery in the puerperium after a neglected perforated appendicitis. A mycotic aneurysm, meaning an infection of the vessel wall which leads to an aneurysmatic dilatation of the vessel, is potentially life threatening if rupture occurs.

Keywords
Aneurysm, embolization, mycotic, puerperium, uterine artery.

Introduction
A mycotic aneurysm is a well-known complication of infectious diseases and sepsis. Aneurysms are associated with significant morbidity and mortality in pregnancy and in puerperium. A pregnant woman presented with unexplained abdominal pain at a term gestation. After an elective induction of labor the patient delivered an uncomplicated healthy child. Three hours after delivery the abdominal complaints increased and the patient developed a hypovolemic shock with signs of intra-abdominal bleeding of unknown origin resulting in a diagnostic laparoscopy. The abdominal cavity showed a hemoperitoneum, a subperitoneal hematoma of the uterus, and a purulent, perforated appendix. The hemoperitoneum and appendix were removed without complications. Two weeks afterward the patient developed abdominal pain again. CT scan showed signs of a leaking mycotic aneurysm in the left uterine artery, which was successfully embolized. We report a successful embolization of a mycotic aneurysm of the left uterine artery in the puerperium.

A mycotic aneurysm is a well-known complication of infectious diseases and sepsis. Bacterial or fungal inflammation of the vessel wall can lead to aneurysmatic dilatation of the vessel and is potentially life threatening if a rupture occurs [1–4]. We report a case of an embolization of a mycotic aneurysm of the left uterine artery 2 weeks after a spontaneous vaginal delivery. Embolization is a well-known method to occlude the uterine artery in hemorrhagia postdelivery or in the treatment of uterine fibroids.

As far as we know, this is the first case report in literature of a mycotic aneurysm of the proximal uterine artery that has been successfully embolized [3, 5–7].

Case Report
A 26-year-old healthy pregnant woman presented with continuous intermittent lower abdominal pain in the left side at 40 + 1 weeks gestation. The pregnancy was uneventful until 36 weeks of gestation, when the woman experienced two short periods of acute left lower abdominal pain, but consulted neither her midwife nor her general practitioner. At 37 weeks a successful external cephalic version was performed.

At admission the woman was tender over the left lower abdomen, but without signs of peritonitis. Her temperature was normal and laboratory examination showed no signs of infection. Because of these unexplained vague
complaints, irregular uterine contractions, and a favorable cervix, it was decided to induce labor by artificial rupture of membranes followed by oxytocin augmentation. The woman had epidural analgesia. After 14 h the patient spontaneously delivered a healthy male infant.

Three hours after delivery the abdominal complaints increased and there were signs of a hypovolemic shock with lowering hematocrit and hemoglobin and transabdominal ultrasound, showed free fluid in the abdominal cavity. Intra-abdominal bleeding was suspected. Diagnostic laparoscopy revealed a hemoperitoneum of 300 mL. After removal of blood and clots, a large subperitoneal hematoma underneath the serosa of the uterus and in the left mesovarium became visible. The appendix was infected, purulent, and possibly perforated since we saw pus in the abdominal cavity. There were multiple adhesions around both adnexae. The appendix was laparoscopically removed and we decided to leave the hemoperitoneum closed, since the size of the subperitoneal hematoma remained stable during surgery. During surgery the patient received a transfusion of three units of packed red blood cells and intravenous cefazolin/metronidazole (3000 mg/1500 mg per day). Due to breastfeeding the antibiotics were switched to amoxicillin/clavulanic acid (1200 mg/4 days) and continued for 10 days. The patient recovered well and was dismissed from the hospital 6 days after surgery.

A pathologic examination confirmed active infection of the appendix. Culture of the appendix showed Escherichia coli and Streptococcus constellatus.

Thirteen days after surgery, the patient was readmitted because of acute left lower abdominal pain and peritoneal tenderness. A transvaginal ultrasound showed a round structure on the left side of the uterus with turbulent (yin-yang sign) Doppler flow (Fig. 1) with minimal fluid in the rectouterine pouch. An abdominal CT scan showed an aneurysm in the left uterine artery. It was a saccular excentric aneurysm with a large retroperitoneal hematoma suggesting perforation. There were no signs of arteriosclerosis. The size and shape of this small based saccular aneurysm suggested mycotic infection of iatrogenic origin. The patient was hemodynamically stable and it was decided to embolize the aneurysm.

The endovascular approach revealed that the bleeding was indeed a result of a large saccular aneurysmatic change in the uterine artery (Fig. 2). This angiography made clear that the intima of the uterine artery vessel was involved. It was possible to bypass the aneurysm and embolize the distal artery to prevent backflow with a 3- and 4-mm Tornado coil. Since this did not result in complete distal embolization, the aneurysmatic lumen was filled with two 20-cm long interlocking detachable coils. The successful embolization of the aneurysm and his secondary bleeding was achieved after placing a final proximal arterial 4-mm Tornado coil (Fig. 2). The patient recovered well and was discharged 2 days after the procedure.

**Discussion**

We describe a successful embolization of a mycotic aneurysm of the left uterine artery in a patient in the puerperium with a neglected, undiagnosed, perforated appendicitis. A mycotic aneurysm is a well-known complication of infectious diseases and sepsis. Bacterial or fungal inflammation of the vessel wall can lead to aneurysmatic dilatation of the vessel and is potentially life threatening if a rupture occurs.

A mycotic aneurysm was first described in 1885 by Sir William Osler, as an infected dilatation of an arterial wall secondary to septic emboli [1, 2].

Nowadays the definition of mycotic aneurysms is based on preexisting arterial disease and/or localized infection
sources. Infection of the artery wall occurs as a result of three possible mechanisms: embolization of bacterial endocarditis, hematogenous spread of micro-organisms in the vasa vasorum (i.e. septicemia), or direct extension from a neighboring source of purulent infection [1, 3]. The vast majority of aneurysm infections result from episodes of septicemia [3]. The sudden appearance of an aneurysm in a septic patient is very suspect of a mycotic aneurysm. Mycotic aneurysms usually lack intimal calcification. In some cases, which probably occurred in our patient, the inflamed appendix became adherent to the wall of the aneurysm resulting in a wall infection [1, 2].

The most common pathogens in mycotic aneurisms are Gram-positive cocci, but Salmonella and other Gram-negative bacteria and also Bacteroides fragilis are described [3]. Among the fungi Candida albicans and Aspergillus are often described as causes of invasive infections [4].

A case report was written in 1999 of a pseudo aneurysm of the uterine artery. Diagnosis was made by duplex Doppler sonography and confirmed by arteriography. It was successfully treated by embolization of the left uterine artery [5]. Preexisting arterial aneurysms in general are associated with an increased risk of rupture in pregnancy. Rupture of these preexisting arterial aneurysms in pregnancy or in puerperium are associated with significant morbidity and mortality. Pregnancy-related rupture of aneurysms of the aorta, cerebral vessels, splenic artery, coronary artery, renal artery, and ovarian artery have been documented. Aneurysms occurring in the uterine artery are rare [3, 5–7].

In the literature, only mycotic aneurysms in the aorta, the internal and external iliacal artery, and femoral artery are described [1, 3, 4].

We suspect that in our case the purulent, perforated appendix caused peritonitis with multiple adhesions around both adnexa and an infection of the wall of the left uterine artery resulting in a mycotic aneurysm.

Clinically diagnosing appendicitis during pregnancy is difficult because of the variable appendiceal position, limited physical examination of the gravid abdomen, and the nonspecificity of symptoms during pregnancy [6]. In several reports, as in our case, diagnosis "mycotic aneurysm" was made after a neglected appendicitis [3, 6]. Earlier in pregnancy our patient experienced this abdominal pain as vague, which was probably due to the appendicitis, peritonitis with secondary the aneurysm of the left uterine artery.

As far as we know, this is the first time that a mycotic aneurysm is described in the uterine artery which was successfully embolized.

Conflict of Interest
None declared.

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