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Rupture of maternal splenic artery aneurysm and fetal demise

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Summary

Splenic artery aneurysm (SAA) is the third most common intra-abdominal aneurysm. This condition, which occurs predominantly in young women, is generally asymptomatic and frequently discovered during pregnancy upon rupture. Reported maternal and fetal mortality are respectively 75% and 72.5-95%. A 40-year-old woman gravida 4 para 3 was referred to the obstetrical emergencies at term for loss of consciousness, nausea, vomiting, and hypotension. At admission, the patient had developed upper abdominal pain. Fetal demise and hemoperitoneum were diagnosed. An abdominal computed tomography (CT) scan revealed SAA rupture. An emergency hemostatic splenectomy was performed followed by a cesarean section with a favorable subsequent outcome. SAA rupture should be considered in the differential diagnosis of acute abdominal pain during pregnancy. Prompt multidisciplinary management is essential for patient's survival.

Key words: Aneurysm rupture; Splenic artery; Pregnancy; Fetal demise.

Introduction

Splenic artery aneurysm (SAA) is the third commonest intra-abdominal aneurysm and the commonest visceral artery aneurysm (60%) [1-4]. This condition is generally asymptomatic and frequently discovered in women of childbearing age (58%) [1, 3, 5].

Because of the paucity of symptoms, the true prevalence of SAA is unknown in general population, but autopsy reports mention a rate of 0.01% to 10.4% [2]. In 69% of cases, it is detected during the third trimester of pregnancy and in 95% of cases upon rupture. Maternal and fetal mortality rates are estimated, respectively, 75% and 72.5-95% [1-4, 6-8]. These high mortality rates are probably explained by the minimal prodromal symptoms, acute deterioration after SAA rupture, and signs that mimic other common obstetric emergencies, such as placental abruption, uterine rupture, amniotic fluid embolization, and in early pregnancies, rupture of an ectopic pregnancy [7, 8].

Case Report

A 40-year-old woman gravida 4 para 3 was referred to the obstetrical emergencies of the Geneva University Hospitals (HUG) at 37 weeks of gestation for loss of consciousness and hypotension. She had presented one episode of brown, non-fecaloïd vomiting. On arrival, she had developed mild abdominal pain and was hemodynamically unstable; her blood pressure was 70/40 mmHg, pulse rate 120 beats/minute. Physical examination revealed a diffuse abdominal pain. There were no uterine contractions or vaginal bleeding and the uterine cervix was closed. She had no signs of preeclampsia, except presence of proteinuria. Fetal heart sounds were absent and fetal demise was confirmed on ultrasound examination. Abdominal ultrasound revealed a massive hemoperitoneum. Hemoglobin level dropped from 120 g/l to 98 g/l with no other alterations in blood tests. Hemodynamic shock was diagnosed and resuscitation was conducted during those investigations with macromolecules and vasoactive amines. Given the patient's hemodynamic shock and the unknown origin of the hemoperitoneum, an abdominal computed tomography (CT) scan was organized and showed a SAA rupture (Figures 1 and 2). An emergency xypho-pubic laparotomy was performed. At peritoneal opening, a hemoperitoneum of three liters was confirmed. A hemostatic splenectomy was performed to stabilize the patient hemodynamically, followed by a cesarean section, with extraction of a stillborn fetus weighing 3,100 g. Pre- and per-operatory resuscitation efforts involved transfusion of eight units of packed red blood cells, seven fresh frozen plasma, cyklokapron and fibrinogen. After splenectomy and weaning from pressors, the patient remained stable. She was transferred to the intensive care unit for 48 hours, with magnesium sulfate administered for the first 24 hours because of suspicion of preeclampsia. Blood pressure and biological markers remained within normal range. Fifteen days after surgery, an infection of a splenic lodge collection was diagnosed. Introduction of empirical antibiotic therapy subsequently adapted to bacterial sampling from cultures (methicillin-sensitive Staphylococcus aureus) allowed the fluid collection to dissipate and the clinical status to improve. The patient was discharged on the 28th postoperative day. She underwent the usual post-splenectomy vaccinations and psychological follow-up was initiated for post-traumatic stress disorder and fetal loss. Histology of the surgical sample confirmed a rupture of a SAA, which was less than two cm, at splenic hilum level.

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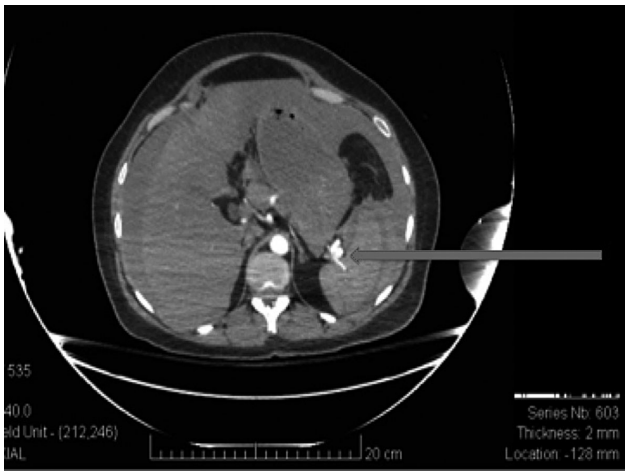

Figure 1. — Injected CT-scan with splenic artery aneurysm which is designated by the arrow.

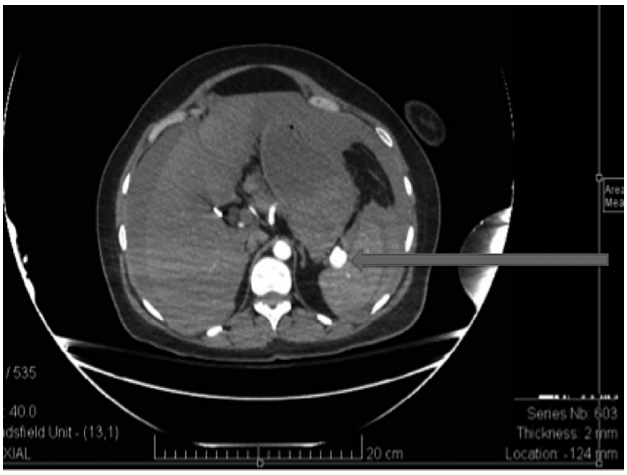

Figure 2. — Injected CT-scan with distal splenic artery aneurysm of 17mm which is designated by the arrow.

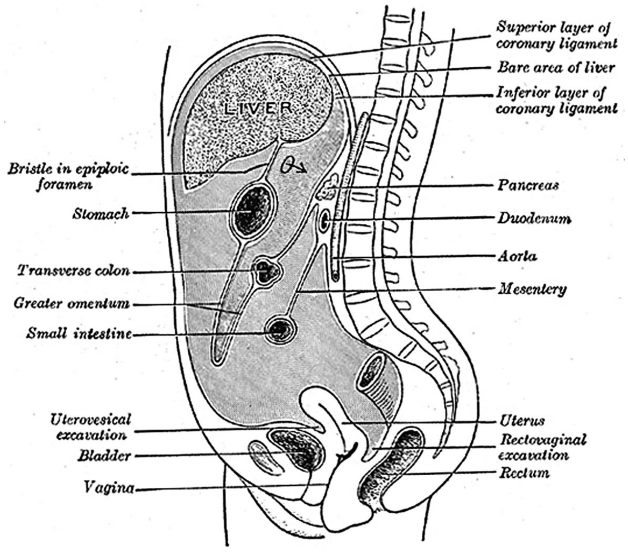

Figure 3. — The greater sac or general cavity (red) and lesser sac or omentum bursa (blue). From 20th U.S. edition of Gray's Anatomy of the Human Body, originally published in 1918.

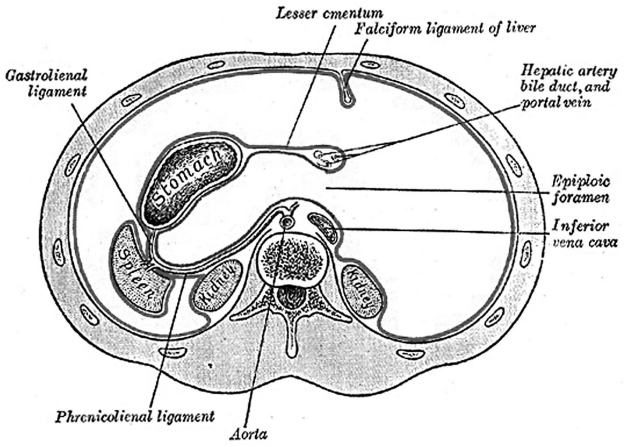

Figure 4. — Horizontal disposition of the peritoneum in the upper part of the abdomen. In red: greater sac end in blue: lesser sac. From 20th U.S. edition of Gray's Anatomy of the Human Body, originally published in 1918.

Discussion

Over 100 cases of SAA rupture during pregnancy have been described. The first recorded case was by Beussier in 1776 and the first favorable maternal issue after treatment was reported by Mac Leod in 1940 [1]. Pregnancy is a known risk factor for SAA rupture. The risk of rupture during pregnancy is estimated at about 20% [6]. Two-thirds of ruptures happen during the third trimester, 13% during the second trimester, 13% per-partum, and 6% in the post-partum period [1, 2, 6, 9]. Prevalence is higher in women suffering from portal hypertension (7%), which increases the risk of rupture earlier on in the pregnancy [6].

During pregnancy, fluctuations in estrogen, and progesterone levels alter arterial wall structure (intimal hyperplasia, interruption of the elastic lamina, and fibrous dysplasia of the media) [1]. Furthermore, splenic artery elasticity is reduced by increased secretion of relaxin hormone in the third trimester [1]. These combined effects stress the vessels wall. Pregnancy is accompanied by hemodynamic changes which add a mechanical component to rupture: increase in cardiac output and blood volume, relative portal congestion, and compression of iliac vessels and the aorta by the gravid uterus, increasing blood pressure in the upper abdomen [1]. The combined effects of these factors are cumulative with each successive pregnancy, increasing not only the incidence of SAA but also the frequency of rupture that is believed to grow as parity rises [1, 2].

A retrospective study by Ha *et al.* examined maternal characteristics and clinical presentations for these SAA. The median age was 28 years, the average number of pregnancies was 2.33, and the mean gestational age at rupture was 34.5 weeks. The average size of aneurysms was 2.25 cm, with more than half of ruptures occurring at aneurysm sizes less than two cm [1]. The most frequent location was the splenic hilum, as in the presented case. Most of the SAAs were discovered upon rupture, with symptoms such as upper abdominal pain (epigastric or left hypochondrium) with dorsal or left shoulder irradiation (Kehr's sign), nausea and vomiting, and hemodynamic instability or shock.

In 20-25% of cases, rupture is preceded by a pre-rupture syndrome: malaise, hypotension, and abdominal pain resolving spontaneously and occurring within 48 hours of aneurysmal rupture [3, 9]. Before admission, the present patient suffered from inaugural malaise without precipitating factor, with loss of consciousness, and no report of abdominal pain at home, upper abdominal pain appearing on arrival at the hospital. This first episode seems to correspond to the aneurysmal pre-rupture syndrome: the aneurysm ruptures in the omental cavity, creating malaise through sudden hypotension and is at first stanched by blood clots or the omentum itself in the lesser sac, blocking the foramen of Winslow, and stabilizing the patient. After a latency period (from minutes to 72 hours), an increase pressure in the lesser sac leads to an intraperitoneal hemorrhage in the large omentum cavity with clinical ascertainment of a hemorrhagic shock (Figures 3 and 4). This phenomenon provides additional time for diagnosis and thus increases the chance of both maternal and fetal survival.

SAA rupture during pregnancy is a surgical emergency requiring prompt multidisciplinary management. Ha *et al.* showed that factors statistically significantly associated with increased maternal mortality were multiple aneurysms, hemodynamic instability, and absence of a general surgeon during laparotomy [1, 3, 6]. Hemodynamic instability and rupture outside of labor were predictive factors of high neonatal mortality, as in the present case.

In the present case, an abdominal CT-scan was performed before surgery in order to determine the location of the bleeding source. Radiological investigations could be considered a waste of time in the emergency of the situation [10], but the patient was still hemodynamically stable. Moreover, the obstetrical team had no reason to suspect uterine rupture or another obstetrical complication, and localizing the true origin of the bleed enabled the authors to justify a multidisciplinary surgical team. This decision is turn allowed for faster control of blood loss, by performing a xypho-pubic laparotomy, and splenectomy before fetal extraction.

Present recommendations differ as to whether propose elective aneurysmal resection surgery or embolization in cases of fortuitous discovery of an SAA over 2-2.5 cm [5, 11] in diameter in women of childbearing age or even under this threshold [12]. If the aneurysm is discovered during pregnancy, the optimal time to operate is in the second trimester, after embryogenesis [1, 13]. Elective surgery can be performed by laparoscopy or laparotomy, and mortality is less than 0.5-1.3% [14].

Conclusion

SAA rupture should be considered as a differential diagnosis in any pregnant woman presenting severe upper abdominal pain or hypovolemic shock. Early recognition and prompt multidisciplinary management are essential for both mother and fetus survival.

References

[1] Ha J.F., Phillips M., Faulkner K.: "Splenic artery aneurysm rupture in pregnancy". *Eur. J. Obstet. Gynecol. Reprod. Biol.*, 2009, *146*, 133.
[2] Sele-Ojeme D.O., Welch C.C.: "Review: Spontaneous rupture of splenic artery aneurysm in pregnancy". *Eur. J. Obstet. Gynecol. Reprod. Biol.*, 2003, *109*, 124.
[3] Wadsworth R., Devine A., Raychaudhuri K.: "Splenic artery aneurysm rupture as a cause of maternal collapse". *Int. J. Obstet. Anesth.*, 1996, *5*, 99.
[4] Cressey D., Reid M.: "Splenic artery aneurysm rupture in pregnancy". *Int. J. Obstet. Anesth.*, 1996, *5*, 103.
[5] Herbeck M., Horbach T., Putzenlechner C., Klein P., Lang W.: "Ruptured splenic artery aneurysm during pregnancy: a rare case with both maternal and fetal survival". *Am. J. Obstet. Gynecol.*, 1999, *181*, 763.
[6] Hillemanns P., Knitza R., Muller-Höcker J.: "Rupture of splenic artery aneurysm in a pregnant patient with portal hypertension". *Am. J. Obstet. Gynecol.*, 1996, *174*, 1665.
[7] Chookun J., Bounes V., Ducassé J.L., Fourcade O.: "Rupture of a splenic artery aneurysm during early pregnancy: a rare and catastrophic event". *Am. J. Emerg. Med.*, 2009, *27*, 898.e5.
[8] Rahmoune F.C., Aya G., Biard M., Belkhayat G., Hamza J., Leperc J., Ouchtati M.: "Splenic artery aneurysm rupture in late pregnancy: a case report and review of the literature". *Ann. Fr. Anesth. Reanim.*, 2011, *30*, 156.
[9] Megevand E., Hirsbrunner P., Fournet N., Francis K., Beguin F.: "Third trimester intra uterine death following spontaneous hemoperitoneum". *Acta Obstet. Gynecol. Scand.*, 1999, *78*, 72.
[10] Khurana J., Spinello I.M.: "Splenic artery aneurysm rupture: a rare but fatal cause for peripartum collapse". *J. Intensive Care Med.,* 2013, *28*, 131.
[11] Mattar S.G., Lumsden A.B.: "The management of splenic artery aneurysms: experience with 23 cases". *Am. J. Surg.*, 1995, *169*, 580.
[12] Samamé J., Kaul A., Garza U., Echeverria A., Galvani C.: "Laparoscopic aneurysm resection and splenectomy for splenic artery aneurysm in the third trimester of pregnancy". *Surg. Endosc.*, 2013, *27*, 2988.
[13] Lang W., Strobel D., Beinder E., Raab M.: "Surgery of a splenic artery aneurysm during pregnancy". *Eur. J. Obstet. Gynecol. Reprod. Biol.*, 2002, *102*, 215.
[14] De Perrot M., Buhler L., Deléaval J., Borisch B., Mentha G., Morel P.: "Management of true aneuysms of the splenic artery". *Am. J. Surg.*, 1998, *175*, 466.

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