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Ruptured splenic artery aneurysm in the 35th week of pregnancy. Medical error or bad luck?

Case report

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Abstract: A 29-year-old woman, para 2 gravida 2, at 35 weeks gestation, came to her local hospital with a sudden onset of severe pain in the left abdomen with associated pain in the back. After 12 hours, the second ultrasonography demonstrated the existence of free fluid in the peritoneal cavity, which was found to be bloody. The surgeon pronounced ruptured spleen the cause of hemorrhage. The autopsy as a direct cause of death revealed massive hemorrhage in the retroperitoneal space and in the peritoneal cavity from a ruptured splenic artery aneurysm. In most cases only double rupture phenomena, in which the hematoma in the lesser sac temporarily tamponades the bleeding aneurysm, is responsible for maternal and fetal survival. In our case, even though the surgeons had about 12 hours to diagnose and treat ruptured the splenic artery aneurysm, they did not succeeded to save the lives of the mother and fetus. Their indifference and delayed surgery intervention cost two lives.

Key Words: aneurysm of splenic artery, rupture, pregnancy, medical error

Splenic artery aneurysm (SAA) was first described on cadavers in 1770 by Beaussier [1]. It was defined as an abnormal dilatation of the splenic artery more than 1 cm in diameter. It predominantly occurs in females, being four times more common and most of the women are pregnant when the lesion is first discovered [2, 3, 4, 5]. The majority of SAAs are <3 cm in diameter and are usually saccular, isolated and located in the mid or distal portion of the splenic artery, frequently at an arterial bifurcation [2, 6, 7].

The precise etiology of splenic aneurysms remains unclear. Different etiological factors attribute to aneurysm formation, including angiodysplasia, portal hypertension, pregnancy, atherosclerosis, diabetes, intracranial aneurysm, polyarteritis nodosa, alpha-1-antitrypsin deficiency and infective factors [4, 8, 9, 10, 11, 12]. Among them, pregnancy has a strong association with splenic artery aneurysm formation [4, 8, 11, 13, 14]. In pregnancy, the influence of hormones, namely estrogen, progesterone and relaxin on the arterial wall, plays a significant role [7, 14, 15].

Ruptured SAA during pregnancy is a rare but

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severe complication. Literature reports a 25% mortality rate for ruptured SAA with an excessive high mortality rate among pregnant women of 75% and an additional fetal mortality rate of even 95% [12, 14, 15, 16, 17]. The high incidence of rupture during the last trimester of 25-45% [18, 19, 20, 21] requires early diagnosis.

A rupture presents with a sudden onset of sharp abdominal pain in the epigastrium or more often left upper quadrant, left shoulder tip pain (Kehr's sign) and hemodynamic instability [12, 15, 22, 23, 24]. This is associated with nausea, vomiting and sudden collapse. Occasionally, ruptures may present with the double rupture phenomena within 48 h, which is present in 20-25% of cases [4, 12, 14, 15, 25]. In this scenario, the first haemorrhage occurs in the lesser sac leading to temporary tamponade. This is followed by flooding through the foramen of Winslow into the peritoneal cavity with resultant severe shock. The double rupture phenomenon was first described by Bockerman in 1930 [2, 6, 7, 12, 15, 26].

In a ruptured SAA, immediate treatment is warranted which can be open, laparoscopic or as embolization of the aneurysm. However, the aim is an immediate resuscitation and cessation of bleeding, which is usually achieved through Caesarian section laparotomy [6, 7, 10, 11, 25, 26, 27, 28].

CASE REPORT

History

A 29-year-old woman, para 2 gravida 2, at 35 weeks gestation, came at 9:05 pm to her local hospital at the Department of Gynecology with a sudden onset of severe pain in the left abdomen with associated pain in the back. She had a pulse rate 90/min, with a blood pressure of 110/70 mmHg on arrival. Physical examination did not reveal any abnormal-pathological changes. Fetal heart tones were registered on the left side, below the navel, 130/min, without contractions of uterus.

Emergency ultrasonography of the abdomen and fetus also did not reveal any abnormal-pathological changes. The abdomen (Morrison), subdiaphragmatic and intrapleural spaces were without free fluid. Ultrasonography showed that pancreas, hilum and the upper pole of the spleen were not seen because they were covered with the gut. The outer contour of the spleen was smooth.

She was given analgesics (metamizole) and she felt better. At 4:15 am, severe pain in the epigastrium occured. Because of the history of gastro-intestinal problems, ranitidine was included in the therapy.

At 8:40 am, first laboratory analysis showed a hemoglobin level of 6.2 g/dL, a hematocrit value of 26.2%, red blood cell 2.96x106/mm^3 and a white blood cell count of 17200/mm^3.

A second ultrasonography, at 9:15 am, demonstrated the existence of free fluid in the peritoneal cavity, which was found to be bloody in the ultrasonographically-guided puncture that followed. Intra-abdominal bleeding was suspected, and the patient was immediately transferred to the operating suite.

After vertical midline laparotomy was performed, the peritoneal cavity was found to be filled with fresh blood and clots, but the wall of the uterus was entirely intact. The surgeon decided to first deliver the fetus by Cesarean section and then treat the hemorrhage. The born infant obtained APGAR score 0 and was pronounced dead. The surgeon noted massive hemorrhage in the left retroperitoneal space and in the greater and lesser omenta. The surgeon pronounced ruptured spleen a cause of hemorrhage, so splenectomy was performed. During surgical procedure, cardiac arrest occured and the following reanimation was unsuccessful. Death pronounced at 11:07am.

Autopsy of woman

The autopsy as a direct cause of death revealed massive hemorrhage in the retroperitoneal space and peritoneal cavity from a ruptured splenic artery aneurysm. The autopsy also revealed that a splenic artery ran directly from aorta (Figure 1), not from the celiac trunk. A medical examiner found a ruptured saccular

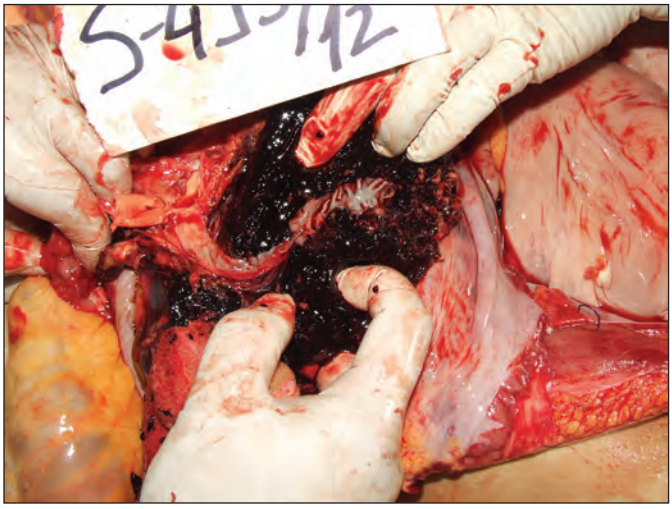

Figure 1. Splenic artery - runs directly from aorta

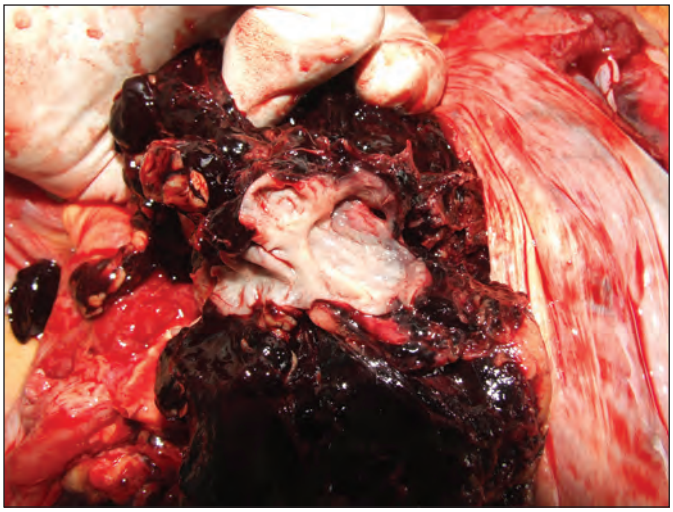

Figure 2. Ruptured saccular aneurysm of the splenic artery

aneurysm on the medial portion of the splenic artery, 2.5 cm in diameter (Figure 2).Examination of uterus and placenta did not reveal any pathological changes.

The spleen was delivered to autopsy in formalin solution. On the visceral surface of the spleen, the medical examiner found a rupture 4x2 cm in diameter and 1.2 cm in depth, but there was no a vital reaction - a rupture without hemorrhage (Figure 3).

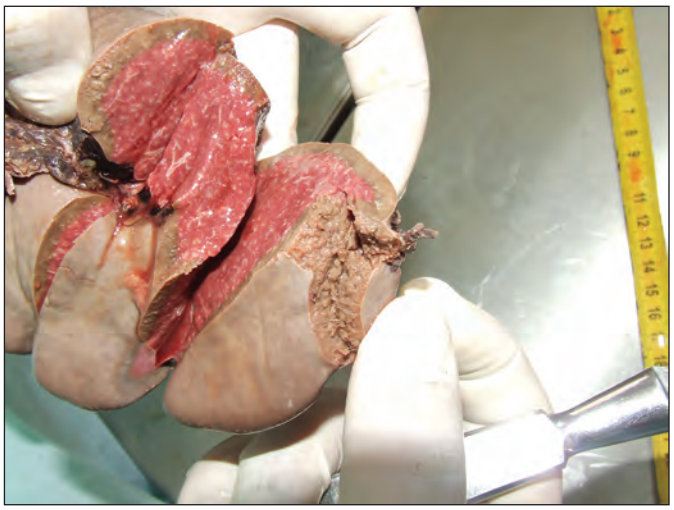

Figure 3. Postmortal splenic rupture

Microscopic findings: Pathohistology examination confirmed a true aneurysm of the splenic artery (Figure 4), and also confirmed that spleen rupture occurred after death.

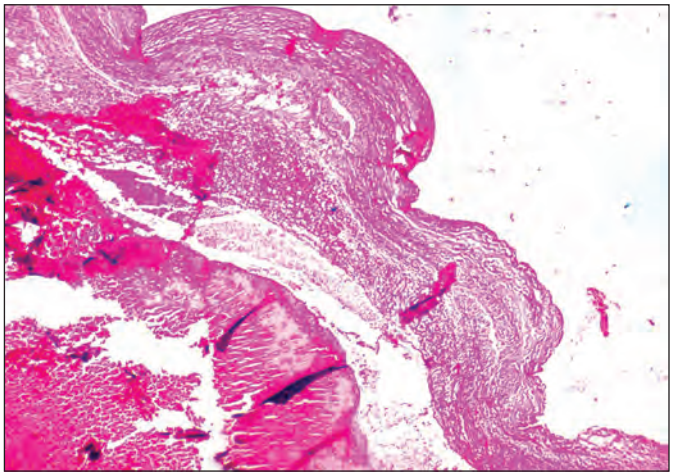

Figure 4. True aneurysm of splenic artery (microscopic view, HE: upper-left part shows normal splenic artery and lower-right part presents thinned muscular layer)

Toxicology findings: Toxicology was negative.

Autopsy of fetus

The autopsy as a direct cause of death revealed an aspiration of the amniotic fluid as a consequence of premature activation of the respiratory center. The fetus weighed 2950 grams; length 49 cm; length from the apex to the plane of the tailbone 34 cm; head circumference 33 cm; chest circumference 29 cm; abdomen circumference 29.5 cm; grain ossification in the lower end of the femur was 5 mm in diameter and in calcaneus was 8x5 mm in diameter. Examination of fetus did not reveal any other pathological changes.

Microscopic findings

Pathohistology examination confirmed an aspiration of the amniotic fluid.

DISCUSSION

Ruptured SAA during pregnancy is a rare event with catastrophic consequences. A diagnosis of ruptured SAA should be considered in any pregnant woman who complains of the sudden onset of severe left upper-abdominal pain regardless of whether pain or shock is prominent at the time of evaluation. This should be kept in mind as a differential diagnosis despite its low incidence because its management requires the engagement of the surgeons from several branches. In literature, rupture of SAA was described as a most commonly during the last trimester of multiparty pregnancy with very high maternal and fetal mortality rate. The pathology causing this association with pregnancy is unclear. The presenting signs and symptoms of splenic artery aneurysm rupture in pregnancy are variable, but most commonly presenting as a non-specific left-side abdominal pain.

The purpose of this report is to describe an unusual case of ruptured SAA during 35th week of pregnancy with a maternal and fetal death. Approximately 25% of SAA ruptures demonstrate the „double rupture" phenomenon.

In most cases only this phenomenon, in which the hematoma in the lesser sac temporarily tamponades the bleeding aneurysm, is responsible for survival, as was the case in our patient. In our case, even this phenomenon gave the surgeons about 12 hours to diagnose and treat ruptured SAA they did not succeeded to save patients and fetus life.

One of the reasons of surgeons failure is unfortunate circumstance that pancreas, hilum and upper pole of spleen, ultrasonography noted, was not seen because they were covered with gut. Second reason of failure is history of gastro-intestinal problems and misinterpreted severe pain in epigastrium. Third reason of failure is 35th week of pregnancy and possible premature birth, even presenting symptoms were telling something else. This was the maior mistake of surgeons. Theirs indifference and delayed surgery intervention (laparotomy) cost 2 lives. Trial is in progress.

In order to prevent cases like this ever happens we present this case. Ruptured SAA should be considered in pregnant women with upper abdominal sequelae especially in multipara. In emergency obstetrics with unknown abdominal bleeding acute or two-stage rupture of an SAA should become a differential diagnosis. Elective surgery is mandatory to prevent serious life-threatening complications from rupture.

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