ELSEVIER

Splenic Artery Rupture During Pregnancy Concealed by a Pancreatic Lymphangioma: A Rare Co-Occurrence

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A pregnant woman presented to a local hospital with abdominal pain and hemorrhagic shock. Emergency caesarean section ruled out an obstetric cause and revealed a large mass, interpreted as a hematoma, with active bleeding of unknown origin. Because of her poor clinical condition, the patient was admitted to our hospital. Computed tomographic findings were suspicious for bleeding originating from the splenic artery. Laparotomy confirmed the presence of a ruptured splenic artery. A splenic artery aneurysm—a relatively well known entity during pregnancy—was absent. Hemostasis was achieved by clipping the artery. A large pancreatic cystic mass, which was misinterpreted earlier as a hematoma, was surgically removed. The pathologic examination revealed a pancreatic lymphangioma, an uncommon benign tumor. The ruptured splenic artery was presumably related to the pancreatic lymphangioma and vascular changes caused by pregnancy. A splenic artery rupture in co-occurrence of a pancreatic lymphangioma is a unique presentation which has not been reported previously.

Severe and prolonged hemorrhagic shock resulting in tissue hypoxia can be life-threatening. Persistent hypoxia can lead to severe complications and death.[1] During pregnancy and in the postpartum period, patients can present with hemorrhagic shock, usually related to gynecologic, obstetric, and/or coagulation disorders.[2–6] However, other less common intra-abdominal causes that necessitate different treatment options should also be considered. In these cases, apart from an obstetrician/gynecologist, the interference of a surgeon, radiologist, and/or intensive care physician is required.

We describe a case of a healthy pregnant woman presenting with acute abdominal pain and severe hemorrhagic shock caused by a ruptured splenic artery (SA). During laparotomy, a large retroperitoneal cyst, which matched the pathologic diagnosis of a pancreatic lymphangioma, was discovered and surgically removed. The rupture was possibly caused by mechanical pressure and traction related to the large lymphangioma combined with vascular changes occurring during pregnancy. Cases of a SA rupture caused by aneurysmatic changes have in particular been described among pregnant women.[4–7] No such aneurysm, however, was observed in our patient. The co-occurrence of both uncommon findings with potential life-threatening consequences has, to our knowledge, never been reported before.

CASE REPORT

A 30-year-old woman, gravida 2 para 1 (37 weeks' pregnant), was transferred from a local hospital and admitted to our hospital because of severe hemorrhagic shock. A few weeks earlier, the patient endured a transient period of abdominal pain for which no obstetric cause was detected. Earlier that day, she underwent a regular obstetric examination, the results of which were

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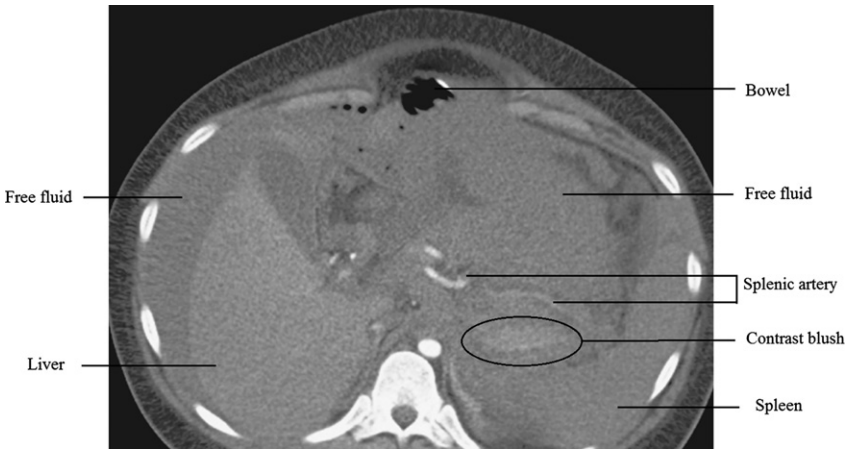

Fig. 1. Contrast-enhanced abdominal computed tomographic angiography (transverse view) showing free fluid around the liver and retroperitoneal space with active contrast blush originating from the splenic artery, representing arterial extravasation.

unremarkable. Later that day, the patient was admitted to a local hospital because of recurrent severe abdominal pain and a loss of consciousness. Medical investigation revealed the presence of shock. Because the fetus appeared to be in distress and a placental abruption was suspected, a caesarean section was emergently performed. Also, a large localized retroperitoneal mass with concurrent active bleeding was observed and interpreted as a hematoma. A placental abruption or uterine rupture was not observed. The origin of the bleeding could not be localized. Because of this, the patient remained hemodynamically unstable despite large volume transfusions and the correction of coagulation disturbances. Despite all efforts, the neonate remained in a poor condition. Both mother and child were transferred to the intensive care units (ICUs) of different large hospitals.

At the time of admission to our hospital, the mother's condition was still life-threatening. She was intubated and mechanically ventilated. The intra-arterial blood pressure was 20/10 mm Hg; she had been hemodynamically stable during transport. The hemoglobin level was 3.3 mmol/L, lactate 7.8 mmol/L in the presence of severe acidosis (pH, 6.77) and persistent coagulation disturbances. A neurologic investigation revealed an eye opening, best motor response, and best verbal response (EMV) score of 1-1-Tube. Subsequent computed tomographic (CT) angiography of the abdomen revealed bleeding that was probably originating from the SA and the giant retroperitoneal cystic mass (18.5 × 16.8 cm) of unknown origin in the left quadrant (Figs. 1 and 2). Radiologic intervention by means of embolization was not considered because of her state of shock and the time needed to prepare for the procedure. Therefore, emergency laparotomy was performed, and afterward the patient was transferred to the ICU. The bleeding was stopped by positioning a clip on the SA, which showed no signs of an aneurysmatic dilatation, and a splenectomy was performed. During laparotomy, the giant tumor seen on the CT scan (Fig. 1) appeared to be a large pancreatic cystic mass. Deroofing of the cyst wall was undertaken. Large quantities of packed red blood cells, fresh frozen plasma units, and platelet concentrate in addition to Ringer's lactate and epinephrine were infused in the pre- and perioperative phase. During surgery, the patient's condition and hemodynamics gradually stabilized. The following day, relaparotomy was performed to remove temporary abdominal packing and to search for active bleeding, which was not noticed. Sedative medication was stopped, whereupon the patient regained consciousness and was successfully extubated. Surprisingly, no neurologic deficits were present. Acidosis was also corrected and inotropic agents were stopped. A histopathologic investigation of the pancreatic mass revealed a benign cyst that matched the diagnosis of a pancreatic lymphangioma ($CD31^+$, $CD34^-$). A malignant cause was excluded. Seven days after laparotomy, the patient was discharged in good clinical condition. Unfortunately, her newborn son did not survive.

DISCUSSION

Defects of the SA are rare and can cause life-threatening situations if a rupture occurs, such as hemorrhagic shock and death. Although uncommon, aneurysms are an important cause of spontaneous ruptures of the SA.[3–5,7] In contrast, a rupture in absence of an aneurysm has rarely been reported. The mortality rate of a ruptured splenic aneurysm is high, ranging from 25–70% depending on its underlying cause. However, its exact pathogenesis remains unclear.[3–5,7] Several congenital and acquired risk factors, like pregnancy, are attributed to its formation. Weakness of the arterial wall and increases in blood pressure are