Rupture of Splenic Artery Aneurysm With Portal Hypertension During Pregnancy: A Case Report

Shahla Chaichian, MD,[1] Abolfazl Mehdizadeh, MD,[2] Abdolrasoul Akbarian, MD,[2] Bahram Groohi, MD,[3] Nazila Khanahmadi, MD,[3] Reza Alaghehbandan, MD[4]

[1] Islamic Azad University, School of Medical Sciences of Tehran, Tehran, Iran

[2] Department of Obstetrics and Gynecology, Iran University of Medical Sciences, Tehran, Iran

[3] Birtle Medical Centre, Assiniboine Regional Health Authority, Birtle MB

[4] Research and Development Division, Centre for Health Information, St. John's NL

Abstract

Background: Spontaneous rupture of a splenic artery aneurysm (SAA) during pregnancy is a rare event with catastrophic consequences. This report presents a case of SAA associated with portal hypertension that ruptured during pregnancy with maternal survival.

Case: A 27-year-old primigravid woman at 31 weeks of gestation presented to the Emergency Department at Pars Hospital in Tehran, Iran with sudden onset of severe abdominal pain. She was in obvious distress with blood pressure of 90/50 mm Hg and a pulse rate of 110 beats per minute. Abdominal ultrasound confirmed free fluid in the peritoneal cavity. The patient was immediately transferred to the operating room. An infant delivered by Caesarean section died shortly thereafter. There was no evidence of placental abruption, but about 2 L of blood was noted in the abdominal cavity. A ruptured SAA was found. Proximal ligation of the splenic artery was performed followed by splenectomy. The patient did well and was discharged on the eighth postoperative day.

Conclusion: This case illustrates the need to consider ruptured SAA as part of differential diagnosis of hemoperitoneum in pregnant women. Immediate surgical intervention is needed to ensure survival of mother and fetus.

Résumé

Contexte : La rupture spontanée d'un anévrisme de l'artère splénique (AAS) au cours de la grossesse constitue un événement rare aux conséquences catastrophiques. Le présent rapport porte sur un cas d'AAS associé à une hypertension portale qui a connu une rupture au cours de la grossesse, sans pour autant entraîner la mort maternelle.

Cas : Une femme primigravide de 27 ans en étant à la 31e semaine de gestation s'est présentée au service des urgences du *Pars Hospital* de Téhéran, en Iran, en raison de l'apparition soudaine de graves douleurs abdominales. Elle présentait une détresse évidente : une tension artérielle de 90/50 mm Hg et un pouls de 110 battements par minute. L'échographie abdominale a confirmé la présence de liquide libre au sein de la cavité péritonéale. La patiente a immédiatement été transférée à la salle d'opération. Le nouveau-né accouché par césarienne est décédé peu après la naissance. Bien qu'aucun signe de décollement placentaire n'ait été constaté, la présence d'environ 2 L de sang a été remarquée dans la cavité abdominale. Une rupture d'AAS a été constatée. Une ligature proximale de l'artère splénique a été effectuée, le tout étant suivi d'une splénectomie. La patiente s'est bien remise et a obtenu l'autorisation de retourner à la maison huit jours à la suite de l'opération.

Conclusion : Ce cas illustre la nécessité d'envisager la rupture d'AAS comme faisant partie du diagnostic différentiel d'hémopéritoine chez les femmes enceintes. Une intervention chirurgicale immédiate est requise pour assurer la survie de la mère et du fœtus.

Key Words: Splenic artery aneurysm, ruptured aneurysm, pregnancy, portal hypertension

Competing Interests: None declared.

Received on October 24, 2005

Accepted on January 11, 2006

J Obstet Gynaecol Can 2006;28(3):302–304

INTRODUCTION

Although rupture of splenic artery aneurysm (SAA) in pregnancy is a rare event, its consequences can be devastating for both mother and fetus. Current estimates of associated maternal and fetal mortality rates are as high as 70% and 90%, respectively.[1,2] Recent reports still show a consistently poor fetal and maternal outcome.[3] Obstetricians and other frontline staff must consider this potentially lethal condition in the differential diagnosis of severe upper abdominal pain in pregnancy if this rate of fatalities per case is to improve. We report a case of maternal survival following SAA rupture during pregnancy.

CASE

A 27-year-old primigravid woman presented at 31 weeks' gestation to the Emergency Department at Pars Hospital in Tehran, Iran. Her pregnancy had been uneventful up to one hour before admission, when she experienced a sudden onset of severe and constant upper abdominal pain that radiated to the back. She also had dyspnea, nausea, and two

episodes of vomiting. There was no vaginal bleeding or rupture of fetal membranes. She had a three-year history of mild epigastric pain with heartburn after meals, and this had not been treated.

On clinical examination, she was in obvious distress (pale with cold peripheries) but not shocked. Her blood pressure was 90/50 mm Hg, and she had pulse rate of 110 beats per minute. The patient had orthostatic hypotension. There was severe tenderness over the uterine fundus. The uterus was soft and no significant uterine contractions were palpable.

Initial blood counts showed a hemoglobin concentration of 74 g/L, normal biochemistry, and no evidence of coagulopathy. Urine analysis was normal. Abdominal ultrasound confirmed free fluid in the peritoneal cavity and enlargement of the spleen.

With the presumptive diagnosis of intraperitoneal hemorrhage, the patient was transferred to the operating room. An urgent laparotomy through a midline incision was made once both obstetric and vascular surgeons were in attendance. The baby was delivered by Caesarean section; at delivery the baby's Apgar score was 6, but the baby died shortly thereafter. There was no evidence of abruptio placentae. An extensive blood clot was found in the lesser sac and across the upper abdomen, and there was bleeding from the splenic artery. Approximately 2 L of blood were removed from the abdominal cavity. The lost blood was replaced with approximately 7 L of intravenous fluid, eight units of packed red cells, and three units of fresh frozen plasma. Although the continuing blood loss made it difficult to determine the original site of the bleeding, a ruptured SAA was found. Proximal ligation of the splenic artery was performed, followed by splenectomy.

After surgery, the patient recovered well and was discharged on the eighth postoperative day. She was referred to medical services for further evaluation. An esophagoscopy revealed severe esophageal varices, and the patient was diagnosed as having portal hypertension. A liver biopsy showed normal histology. Three months later, a repeat esophagoscopy showed no sign of esophageal varices. The patient had a pregnancy with an uncomplicated delivery one year later.

DISCUSSION

The principal complication of an SAA is rupture, and the reported risk varies from 3% to 9.6%.[4,5] Rupture occurs more frequently during pregnancy, when reported rates range from 20% to 50%.[5–7] Although rupture can occur at any time during pregnancy and during the immediate postpartum period, 69% of cases are found in the third trimester.[4] Rupture during pregnancy is associated with a maternal mortality rate of 70% and a fetal mortality rate of 90%.[1]

The literature now contains more than 100 cases of ruptured SAAs in pregnancy but only 16 cases of maternal and fetal survival.[8–10] Although the average parity of women at rupture is 4.5,[4] nulliparous women are not immune, as demonstrated by this case and others.[11,12]

In most cases, the cause of an SAA is unknown. However, whatever the etiology, the ultimate pathology is local failure of the connective tissue of the arterial wall to maintain the integrity of the vessel.[8] Potential risk factors include portal hypertension, congenital abnormalities of the vessels, inherited vascular and connective tissue disorders, vascular trauma, inflammatory processes, and degenerative arterial disease.[6,8] Portal hypertension may have been a risk factor in the present case.

Generally, SAA manifests clinically in one of three ways: (1) as an incidental finding; (2) with abdominal pain of varied severity (which was the clinical manifestation of the present case); or (3) after rupture, with hypotension and collapse. The predominant presenting symptom is pain, which can be acute or insidious in onset and continuous or intermittent. It is located mostly in the left flank or left upper abdomen. It can also be located in the epigastric region or right upper abdomen.[4] The pain is usually described as sharp or searing and radiates to the back, to the tip of the left shoulder (Kehrs' sign), or into the legs.[6,13–15] There may be associated nausea and vomiting.[16] When rupture occurs, the pain is acute and may last from hours to days.[17]

There are no physical signs that reliably indicate the presence of an SAA.[18] In the early stages of rupture, diffuse tenderness in the upper abdomen or over the uterine fundus may be elicited. In severe cases, and with significant blood loss, signs of peritoneal irritation, hypotension, and shock are present.

Rupture of SAA in pregnancy is difficult to diagnose because it shares signs and symptoms with other conditions, such as placental abruption, uterine rupture, perforated ulcer, or rupture of other arterial aneurysms.[11, 19–21] Often the diagnosis is made only at surgery.[1,2,11] In a suspected unruptured SAA the gold standard for diagnosis is arteriography,[22] although ultrasonography and pulsed Doppler are preferable in pregnancy.[7] Computed tomography is also useful.[1,18] When a patient with a ruptured SAA presents with acute abdominal pain, an emergency ultrasound scan may reveal free fluid in the upper abdomen, and the diagnosis is subsequently confirmed at laparotomy.[1,20]

When there is a rupture, management decisions are clear-cut and uncontroversial. Resuscitation and arrest of hemorrhage by emergency surgery are essential for the

survival of both mother and fetus. Splenectomy is often performed in addition to resection of the aneurysm.[7,22] To ensure the best surgical management for the patient, the obstetrician must involve a general or vascular surgeon as soon as a ruptured SAA is suspected.

CONCLUSION

A diagnosis of ruptured SAA should be considered in any pregnant woman who presents with severe left upper abdominal pain or hypovolemic shock. It is essential that obstetricians are alert to the prodromal and catastrophic symptoms of SAA. A high index of suspicion, early recognition, and prompt management, including early involvement of a general surgeon, are vital to the survival of mother and fetus. An asymptomatic SAA should be resected electively when found in a pregnant woman or in a woman of childbearing age.

ACKNOWLEDGEMENT

The woman whose story is told in this case report has provided signed permission for its publication.

REFERENCES

1. Hillemanns P, Knitza R, Muller-Hocker J. Rupture of splenic artery aneurysm in a pregnant patient with portal hypertension. Am J Obstet Gynecol 1996;174:1665–6.
2. Fender GRK, Haslett E, Leary T, Bland E, Hackett G. Management of splenic artery aneurysm rupture during trial of scar with epidural analgesia. Am J Obstet Gynecol 1999;180:1038–9.
3. Shahabi S, Jani J, Masters L, Cobin L, Greindl J. Spontaneous rupture of a splenic artery aneurysm in pregnancy: report of two cases. Acta Chir Belg 2000;100:231–3.
4. Trastek VF, Pairolero FC, Joyce JW, Hollier LH, Bernatz PE. Splenic artery aneurysms. Surgery 1982;91:694–9.
5. Salo JA, Salmenkivi K, Tenhunen A, Kivilaakso EO. Rupture of splanchnic artery aneurysms. World J Surg 1986;10:123–7.
6. Messina LM, Shanley CJ. Visceral artery aneurysms. Surg Clin North Am 1997;77:425–42.
7. Lang W, Strobel D, Beinder E, Raab M. Surgery of a splenic aneurysm during pregnancy. Eur J Obstet Gynecol 2002;102:215–6.
8. Selo-Ojeme DO, Welch CC. Spontaneous rupture of splenic artery in pregnancy—review. Eur J Obstet Gynecol Reprod Biol 2003;109, 124–7.
9. Al Asfar F, Saber M, Dhar PM, Al Awadhi N. Rupture of splenic artery aneurysm during labor: a case report of maternal and fetal survival. Med Princ Pract 2005;14(1):53–4.
10. Reches A, Almog R, Pauzner D, Almog B, Levin I. Spontaneous splenic rupture in pregnancy after heparin treatment. BJOG 2005;112(6):837–8.
11. Caillouette JC, Merchant EB. Ruptured splenic artery aneurysm in pregnancy. Twelfth reported case with maternal and fetal survival. Am J Obstet Gynecol 1993;168:1810–3.
12. Czekelius P, Deichert L, Gesenhues T, Schulz KD. Rupture of an aneurysm of the splenic artery and pregnancy: a case report. Eur J Obstet Gynecol Reprod Biol 1990;38:229–32.
13. Hallett JW. Splenic artery aneurysms. Semin Vasc Surg 1995;8:321–6.
14. Prabulos AM, Chen HH, Rodis JF, Ruby S, Campbell WA. Angiographic embolization of ruptured renal artery aneurysm during pregnancy. Obstet Gynecol 1997;90:663–5.
15. Chen CW, Chen CP, Wang KG. Splenic artery aneurysm rupture in the second trimester. Int J Gynecol Obstet 1995;49:199–200.
16. Holdsworth RJ, Gunn A. Ruptured splenic artery aneurysm in pregnancy: a review. Br J Obstet Gynaecol 1992;99:595–7.
17. de Perrot M, Buhler L, Deleaval J, Borisch B, Mentha G, Morel P. Management of true aneurysms of the splenic artery. Am J Surg 1998;175:466–8.
18. Mattar SG, Lumsden AB. The management of splenic artery aneurysms: experience with 23 cases. Am J Surg 1995;169:580–4.
19. Czekelius P, Deichert L, Gesenhues T, Schulz KD. Rupture of an aneurysm of the splenic artery and pregnancy: a case report. Eur J Obstet Gynecol 1991;38:229–32.
20. 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;181:763–4.
21. Lovsin B, Ravnikar J, Guzej Z. Splenic artery aneurysm. Am J Obstet Gynecol 2000;183:781.
22. Wagner WH, Allins AD, Treiman RL, Cohen JL, Foran RF, Levin PM, et al. Ruptured visceral artery aneurysms. Ann Vasc Surg 1997;11:342–7.