

CASE REPORT

Fatal spontaneous rupture of splenic artery aneurysm in third trimester pregnancy

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Abstract

Introduction: Unforeseen emergency in late pregnancy can be catastrophic and cause unexpected maternal and foetal demise. Moreover, lack of awareness and failure of prompt treatment raise mortality rate. Such fatalities warrant a forensic autopsy as it may raise redundant medico-legal concerns. **Case Report:** We report a case that revealed significant intra-abdominal haemorrhage at autopsy. The source of haemorrhage was at the spleen hilum and histology established rupture of splenic artery aneurysm. There was no associated obstetric cause found. **Conclusion:** Knowledge of spontaneous rupture of splenic artery aneurysm in late pregnancy is essential for monitoring maternal and foetal, morbidity and mortality. However, in the eventuality of death a comprehensive forensic autopsy is the only investigation to recognise such calamity and clear clinical confusion.

Keywords: Spontaneous rupture, splenic artery aneurysm, late pregnancy, haemoperitoneum, forensic autopsy

INTRODUCTION

Abdominal pain in 3rd trimester pregnancy can be of obstetric or non-obstetric origin.¹⁻³ A list of conditions is listed in literature and amongst these spontaneous ruptures of spleen and splenic vessels are documented.^{4,5} Despite this, sudden death due to asymptomatic and non-traumatic rupture of splenic artery in third trimester pregnancy is extremely rare and carries high maternal and foetal morbidity and mortality.¹⁻⁹ Such fatalities were predominantly attributed to difficulty in diagnoses due to lack of awareness. A fatal outcome requires a comprehensive forensic autopsy investigation to conclude precise cause of death and answer next-of-kin concerns.^{5,6}

CASE REPORT

A 29-year-old primigravida in 36 weeks of gestation complained of sudden onset abdominal pain in midst of baby shower. Mistaking the pain as labour contractions, she was immediately transported to hospital. However, she collapsed and could not be revived. The foetal heart sounds were not recordable. She was on regular iron and

folate supplements and an ante-natal check-up a day prior was uneventful. There was no history of medical illness or allergies. The social habits did not reveal anything unusual. No clinical cause for death was identified therefore a medico-legal autopsy was warranted.

An autopsy was conducted on same day. Initial observation revealed a young gravid female weighing 65 kg and 159 cm in height. There was no evidence of external injuries or bleeding per vagina however, generalised pallor was noted. Peritoneal cavity contained 2000 mL of blood-stained fluid and blood clot. Additional 720 gm haematoma was noted originating from splenic hilum extending further along length of pancreas (Fig. 1). The spleen (Fig. 1A) weighed 170 gm and its cut surface showed diffuse parenchymal haemorrhage (Fig. 1B). The splenic artery (Fig. 1C) was dilated close to hilum and measured 16 mm in diameter using a digital Vernier calliper. The remainder splenic artery and adjacent blood vessels of abdomen did not reveal any abnormality. Pancreas tail (Fig. 1D) was engulfed with haemorrhage and weighed 255 gm. The left adrenal gland was surrounded



FIG. 1: Specimen of spleen and pancreas showing (A) spleen, (B) haemorrhage in spleen parenchyma, (C) dilated splenic artery and (D) pancreas tail.

by haematoma and weighed 15 gm. Liver and kidneys appeared pale. The brain and heart showed normal configuration. Both lungs were pale and there was no evidence of pulmonary thromboembolism.

The gravid uterus measured 40 x 35 cm and did not show any lacerations. A 2.4 kg lifeless male foetus in cephalic presentation was retrieved. The general measurements of the foetus were: crown-heel length 51 cm, crown-rump length

33 cm and foot length 7 cm consistent with 36 weeks gestation. No congenital anomaly was noted. Placenta was normal measuring 19 x 13 x 2 cm and located in the upper uterine segment. The cord length was 48 x 1.5 cm with 3 blood vessels.

Histology of the splenic artery disclosed aneurysmal dilatation with focal area of discontinuity and thinning of the arterial wall (Fig. 2). The arterial wall also exhibited marked

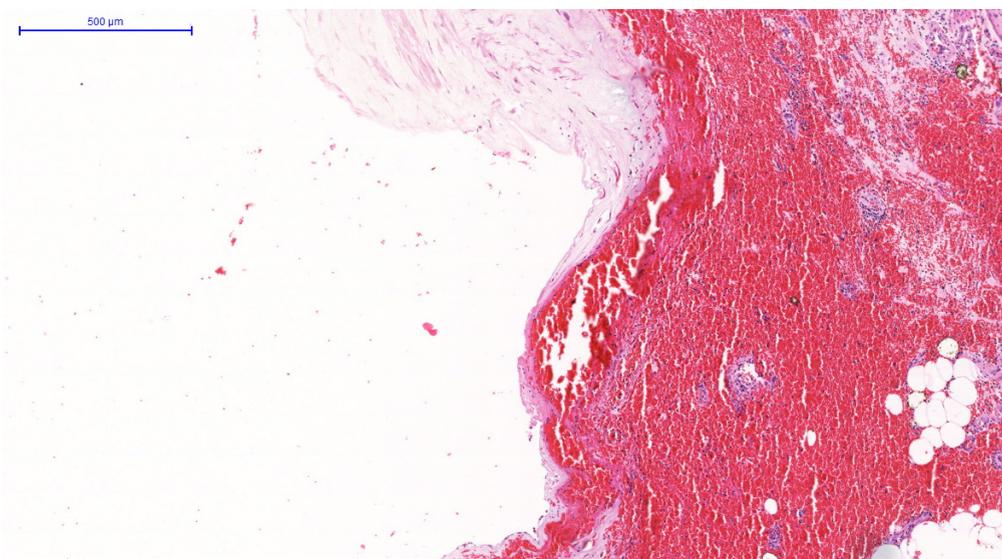


FIG. 2: Aneurysmal dilatation of splenic artery with focal area of discontinuity and thinning of the arterial wall (H&E, x4).

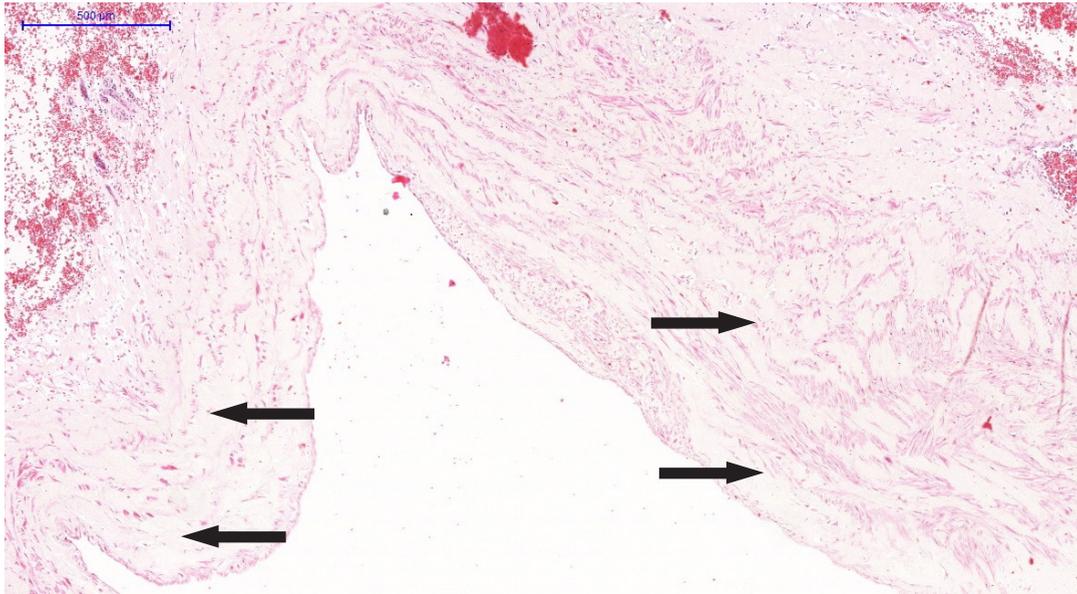


FIG. 3: Thickened portion of splenic artery exhibiting marked mediolysis of muscular and elastic fibres with interspersed pools of mucoid material (arrow) (H&E, x4).

mediolysis of muscular and elastic fibres with interspersed pools of mucoid material (Fig. 3). In addition, fibroblastic proliferation of intima, acute inflammatory infiltrate and adjoining recent haemorrhage was noted. Special stain Verhoeff's Van Geison revealed irregular remnants of fragmented elastin fibres (Fig. 4). Histology of rest of spleen parenchyma showed haemorrhage.

The splenic vein was intact. The lungs showed aspirated food material a complication of long-standing resuscitation. There was no evidence of amniotic fluid embolism. The other organs did not reveal any pathology. Toxicology analysis report was negative for alcohol and common drugs.

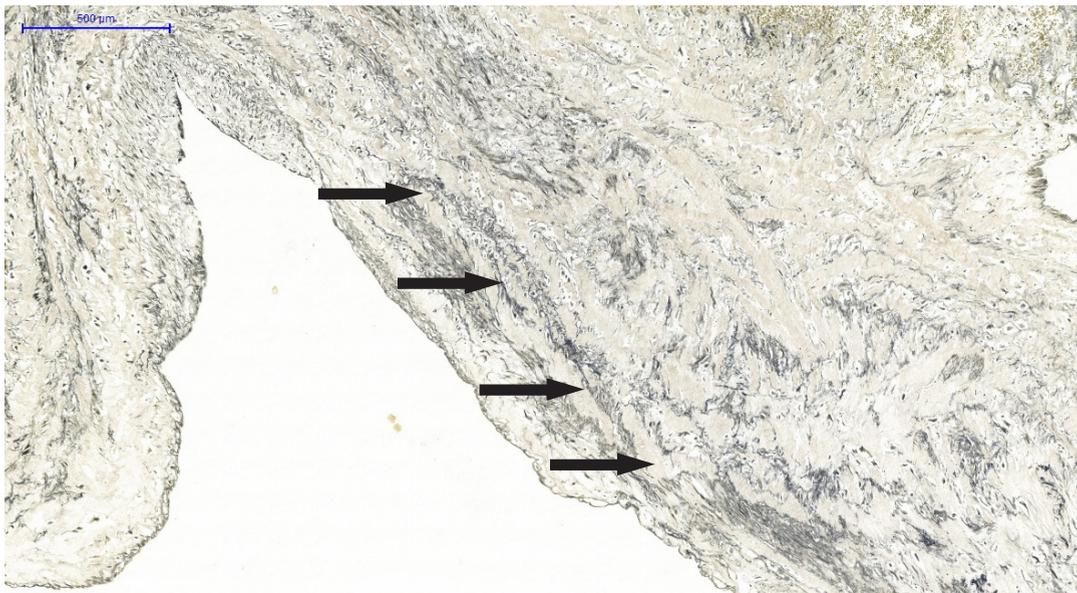


FIG. 4: The same segment of the splenic artery showing irregular remnants of fragmented elastin fibres (arrow) (Verhoeff's Van Geison (EVG), x4).

DISCUSSION

Splenic artery aneurysm (SAA) was first described on cadavers by Beaussier in 1770.¹⁰ It is the most common visceral aneurysm and third most common intra-abdominal aneurysm after the aortic and common iliac artery aneurysm. Clinically it has a higher incidence in women with a two-to-five-fold increase over men.⁹ Spontaneous haemorrhage from splenic artery aneurysm (SAA) rupture is a rare and serious complication during pregnancy.¹⁻⁹ In women the incidence of visceral aneurysms mounts with pregnancy and parity.⁹ The greater preponderance of females in surgical cases is accounted for by the frequency of rupture of aneurysm occurring during pregnancy which in no doubt was due to the increased intra-abdominal pressure during last trimester.⁹

We presented a case where the diagnosis of spontaneous rupture of splenic artery aneurysm in late pregnancy was diagnosed at post mortem.^{5,6,9} Similar to our case, the mother usually presents in shock, and, it is not unusual for emergency physicians to look for obstetric cause.^{1,2,4,7} This however, could lead to delay in diagnosing non-obstetric causes, raising the morbidity and mortality for mother and foetus.^{1,5}

A variety of obstetric and non-obstetric causes account for acute abdomen in pregnancy. The obstetric causes are managed promptly. It is the rare non-obstetric (0.23%) conditions specific to pregnancy where timely diagnosis may prove lifesaving.^{2,3,8} Though arteriosclerosis, hypertension and increased intra-abdominal pressure are considered frequent causes of aneurysmal rupture, other factors, such as, congenital, mycotic, arterial degeneration, atheroma, embolism, pregnancy, trauma are also recognised.⁹ Further, spontaneous bleeding during pregnancy can also arise from utero-ovarian, splenic, hepatic, superior mesenteric or celiac vessels. In principle arterial bleeds are copious causing quick cardiovascular collapse and may present with features of hypovolemic shock.^{3,7}

A variety of factors are associated to such a presentation. However, rupture of SAA during pregnancy remains uncommon. Moreover, the presentation is strikingly similar not only to routine obstetric causes, but also other rare non-obstetric entities encountered during pregnancy notably spontaneous ruptures of spleen, splenic vein and utero-ovarian vessels.¹¹⁻¹⁴ Though the exact mechanism is still not clearly

understood, it is noteworthy to understand that the cardiovascular system undergoes significant anatomic and physiologic modification owing to hormonal surge during pregnancy.^{12,15}

Additionally, pregnancy itself acts as nature's stress test causing increased blood flow and changes in vascular wall. Besides, hormonal, genetic, thrombotic and mechanical forces increase the risk of ruptures through weakness in vessel wall.¹² Oestrogen and progesterone alter and weakens the arterial wall by causing medial degeneration. Relaxin is also thought to affect the elasticity of the vessels through altered collagen and elastin deposition.¹⁶ The histology findings in our case clearly demonstrate such arterial wall disruption with aneurysmal dilatation and rupture.

Though such aneurysms are encountered commonly in clinical practice and indicate surgical exploration, the autopsy incidence of aneurysm of the splenic artery varies from 0.039% to 0.8% and is dependent predominantly on the awareness and thoroughness of examination.^{1,2,5,9} A search through our records established this as the first case where mother and foetus succumbed to spontaneous rupture of splenic artery aneurysm in third trimester pregnancy. As of now few cases of splenic artery aneurysmal rupture during pregnancy are recorded at autopsy. Dearth of cases is the root cause for delayed diagnosis causing high mortality. Therefore, this condition remains obscure and unpreventable.

Clearly, these cases are a clinical scenario that overlaps specialties due to variety of probable causes. Thus, the obstetrician/gynaecologist should obtain added assistance from surgical and medical specialist as the approach is like that for non-pregnant patients with acute abdomen. Non-existent obstetric complication should be an indication for laparotomy which might prove lifesaving. Moreover, awareness at antenatal check-up is recommended for prevention, early diagnosis and intervention. In unfortunate circumstances, a comprehensive forensic autopsy investigation is mandatory.

CONCLUSION

Spontaneous rupture of splenic artery aneurysm in pregnancy is usually misdiagnosed because the "eyes do not see what the mind does not know".¹⁷ Nevertheless, any sudden-onset abdominal pain in pregnancy should raise suspicion of spontaneous abdominal bleed as rapid diagnosis is essential in view of hypovolemic shock.

Aggressive volume replacement and prompt surgical intervention are favourable. In addition, ante-natal check-up should involve pre-cautious radiological screening of abdominal visceral vessels.¹⁸ In cases of unexpected death, a comprehensive forensic autopsy is mandatory.

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