

# Successful management of an unusual presentation of ruptured splenic artery aneurysm in the third trimester presenting as right sided abdominal pain. A case report

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## Abstract

Spontaneous rupture of a splenic artery aneurysm in pregnancy is a rare cause of major obstetric haemorrhage. It carries a high mortality rate of up to 75% and foetal mortality of 95%. Presentation of this condition during pregnancy can be highly variable and a high index of suspicion is required to diagnose this condition. Prompt multidisciplinary management along with aggressive resuscitation is vital for a positive outcome. We report a case of successful management of an unusual presentation of a ruptured splenic artery aneurysm in the third trimester presenting with right sided abdominal pain with a good maternal and foetal outcome and aim to present a synopsis of management of this condition.

## INTRODUCTION

The true prevalence of splenic artery aneurysm (SAA) in pregnancy is yet unknown as majority of the cases remain asymptomatic. It carries a mortality of 25 % in non-pregnant, which increases to 75% in pregnancy with foetal mortality of 95%.<sup>1</sup> The risk of rupture is about 25% - 45% in the last trimester.<sup>2</sup> Four maternal deaths were reported in the U.K due to SAA rupture in the Confidential Enquiry into Maternal and Child health (CEMACH) report (Saving Mothers Lives 2003-2005); while between 1988 and 1999, there were seven maternal deaths. Till date less than 20 cases of both maternal and foetal survival have been reported in medical literature.

## CASE REPORT

A 37 year old, G3 P2, ASA I, Caucasian female was admitted to the maternity ward with ten day history of intermittent sharp right upper quadrant pain radiating to the right arm and shoulder which suddenly increased in severity at 31 weeks gestation. She was otherwise asymptomatic. Her past medical history included previous two normal vaginal deliveries. During this pregnancy, she was under antenatal surveillance for oligohydramnios and intrauterine growth retardation (IUGR) with regular ultrasound scans. Previous ultrasound scan done at 29 weeks gestation showed a normal foetus and placenta with reduced liquor. At 30 weeks

gestation, she reported intermittent episodes of sharp pain and tenderness in the right upper quadrant radiating to the right shoulder and arm. A thorough physical examination, ultrasound scan, blood tests, and routine observations were normal so she was discharged with a follow up appointment.

On examination, her arterial blood pressure was 123/80 mm Hg. Heart rate was 108 beats min<sup>-1</sup>. She was afebrile and had oxygen saturation of 99% on room air. No pallor or icterus was noted. Cardio respiratory and neurological examination was normal. She had tenderness and guarding in the right hypochondrium. Vaginal examination was normal and the uterus was nontender. CTG showed a normal reactive trace.. An urgent surgical review was requested, as no obvious obstetric pathology was evident. The senior consultant surgeon on call reviewed the patient and a provisional differential diagnosis of biliary colic was considered. Blood results obtained showed Hb 10.7 g dL<sup>-1</sup>, white cell count  $10 \times 10^9 \text{ L}^{-1}$  with normal clotting, renal and liver function tests.(see table 1). Intravenous fluids and broad-spectrum antibiotic (cefuroxime 1.5gm) were prescribed. A repeat ultrasound scan of the abdomen and pelvis was planned. She was reviewed again by the surgical team by which time her pain had completely settled. The following morning (about 22 hours into her initial admission) as she was being moved for the ultrasound scan, she suddenly developed severe left flank pain and felt dizzy.

On examination, she was profoundly hypotensive with a blood pressure of 80/44 mm Hg and a prolonged capillary refill time. Heart rate was 122 beats min<sup>-1</sup>. She was tachypnoeic and S<sub>po2</sub> was 94% on air. Marked mucosal pallor was noted. She had generalised abdominal guarding, rigidity and rebound tenderness. Uterus was noted to be firm with some suprapubic tenderness. CTG and vaginal examination were normal. Additional intravenous access was obtained with 14G cannulae. High flow oxygen was administered and fluid resuscitation commenced with i.v colloids (Gelofusine). Blood was requested. Suspecting a possible uterine rupture decision was taken to perform an emergency laparotomy and caesarean section. 30 mls of 0.3 M Sodium citrate was administered along with i.v. ranitidine 50 mg and i.v. metoclopramide 10 mg. Despite aggressive fluid resuscitation with O negative blood (4 units) and i.v colloid (Gelofusine), patient rapidly developed hypovolemic shock.

Upon arrival into the operating theatre, routine monitoring was commenced. Patient was drowsy with a heart rate of 162 beats min<sup>-1</sup>. Her arterial pressure was 68/ 30 mm Hg. S<sub>po2</sub> was difficult to register. Pre oxygenation was commenced immediately and a rapid sequence induction was performed with a careful titration of etomidate 10 mg, midazolam 5 mg. Suxamethonium 100 mg was given to facilitate orotracheal intubation with a 7.5 mm cuffed endotracheal tube. Anaesthesia was maintained with oxygen (F<sub>Io2</sub> 1.0), sevoflurane (0.5 MAC) and 50 mcg boluses of fentanyl. Neuromuscular blockade was maintained with i.v. atracurium. Lungs were ventilated artificially with intermittent positive pressure ventilation mode (Dräger, Cato) with F<sub>io2</sub> of 1.00, tidal volume of 550 mls, and a respiratory frequency of 10-12 breaths min<sup>-1</sup> on a circle system. Rapid transfusion was commenced with packed red blood cells (PRBC) through two peripheral large bore cannulae (14G) using pressurised bags. During and after induction patient was severely hypotensive with a barely palpable peripheral pulse; therefore, we were unable to measure non-invasive blood pressure accurately. Multiple attempts at securing an arterial line initially were unsuccessful. Patient required multiple 10 µg boluses of i.v. adrenaline (1:100,000) to maintain adequate perfusion. After multiple unsuccessful attempts, the right radial artery was cannulated. Her arterial pressure at this point was 72/40 mm Hg. A noradrenaline infusion was commenced at 0.3 µg kg<sup>-1</sup> min<sup>-1</sup> and titrated to achieve a mean arterial pressure (MAP) between 50-60 mm Hg. Right internal jugular central venous

access was obtained and was transduced to monitor central venous pressure. Initial CVP reading was 3 mm Hg. Initially 10 units of packed red blood cells and 4 units of fresh frozen plasma (FFP) were transfused through a fluid warmer. A blood sample was analysed for clotting studies and platelet count. It showed a prothrombin time (PT) of 13.4s, platelet count of 49 x10<sup>9</sup> L<sup>-1</sup>, fibrinogen level of 1.32 g l<sup>-1</sup>, activated partial thromboplastin time (aPTT) of 40s with an aPTT ratio of 1.6. Arterial blood gas (F<sub>io2</sub> 1.0) showed pH 7.26, P<sub>co2</sub> 4.8 kPa. P<sub>o2</sub> 35.86 kPa, bicarbonate 16.7 mmols L<sup>-1</sup> and base excess of -10.9 mmols L<sup>-1</sup>. Haemoglobin was checked using the HemoCue™ Haemoglobin analyser (HemoCue Ltd, Sheffield. UK) and was found to be 4 g dL<sup>-1</sup>. Further packed red blood cells (10 units) and FFP (4units) were transfused. Pooled platelets and cryoprecipitate were also transfused during the course of the operation guided by clotting studies. Blood glucose, calcium and potassium levels were monitored with regular arterial blood gas samples. An upper body forced air warmer (Bair Hugger®, Arizant UK, Wakefield, West Yorkshire, UK) was applied to keep the patient warm. Core temperature was monitored using an oesophageal temperature probe (Temprecise™, Arizant UK, Wakefield, West Yorkshire. UK). A remifentanyl (0.2 µg kg<sup>-1</sup>.min<sup>-1</sup>) infusion was started after the baby was delivered.

Surgery was commenced immediately after endotracheal intubation and a massive hemoperitoneum was noted (estimated blood >3 litres) with an intact uterus. A live female infant was immediately delivered by lower segmental caesarean section. Baby was handed over to the paediatric team, who immediately intubated the infant and transferred it to the special care baby unit. The Apgar scores were 2 and 5 at 1 and 5 minutes. Uterus was sutured and simultaneously incision was extended up to the xiphisternum, Torrential haemorrhage was noted coming from behind the stomach near the superior border of spleen. Suspecting a ruptured splenic artery aneurysm, the lesser sac was opened. A large retroperitoneal haematoma made it impossible for the exact bleeding point to be visualised. After much difficulty temporary haemostasis was achieved with 2/0 vicryl sutures applied to the retroperitoneal tissue. However, the proximal end of the splenic artery could not be visualised because of the tissue oedema and haematoma. Therefore, specialist hepatobiliary pancreatic surgical input was requested from the regional specialist centre. Hepatobiliary team arrived in an hour and after further dissection managed to ligate the proximal end of the splenic artery. Floseal™ (Baxter, U.S.)

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haemostatic matrix was applied to the vascular bed. Spleen was left intact and abdomen was closed with PDS sutures.

The estimated blood loss during the procedure was over 16 litres. In total, the patient required 20 units of packed red blood cells, 8 units of FFP, 2 units of cryoprecipitate, and 2 pools of platelets in addition to 2 litres of crystalloids (Hartmann's solution). Sedation was commenced with propofol 1%, 2 mg kg<sup>-1</sup> hr<sup>-1</sup> and i.v. alfentanil infusion at 0.5- 1.0 µg kg<sup>-1</sup> min<sup>-1</sup>. Inotropic support was discontinued, as the arterial pressure was 130mm Hg systolic and 60mm Hg diastolic with a CVP of 10 mm Hg. Core body temperature was 35° Celsius. Arterial blood gas showed a pH 7.28, P<sub>O2</sub> 14 kPa, P<sub>CO2</sub> 6.1 kPa, bicarbonate 22 mmols L<sup>-1</sup> and base excess of -6 mmols L<sup>-1</sup>. Clotting studies repeated post operatively were normal and Haemoglobin was 9.3 g dL<sup>-1</sup> with platelet count of 70 x 10<sup>9</sup> L<sup>-1</sup> (see table 1).

Ventilation was continued and patient was transferred to intensive care unit. Following morning, artificial ventilation was discontinued and tracheal tube was removed. She underwent a contrast CT of abdomen and pelvis followed by a mesenteric angiography, which showed no sign of any other aneurysms or leak. Patient and her baby made an uneventful recovery and both were subsequently discharged.

### Figure 1

Table 1

|                      | Units                              | Ref. range  | Day 1 | Day2 preop | Post op |
|----------------------|------------------------------------|-------------|-------|------------|---------|
| Haemoglobin          | g dL <sup>-1</sup>                 | 11.5 - 16.5 | 10.7  | 7.7        | 9.3     |
| White cell count     | X 10 <sup>9</sup> L <sup>-1</sup>  | 4.0 - 11.0  | 10.0  | 11.0       | 4.3     |
| Platelets            | X 10 <sup>9</sup> L <sup>-1</sup>  | 150 - 450   | 170   | 124        | 70      |
| Red cell count       | X 10 <sup>12</sup> L <sup>-1</sup> | 3.8 - 4.8   | 3.32  | 2.37       | 3.12    |
| Hematocrit           |                                    | 0.37 - 0.47 | 0.326 | 0.235      | 0.28    |
| Neutrophil count     | X 10 <sup>9</sup> L <sup>-1</sup>  | 2.0 - 7.5   | 5.9   | 8.6        | 3.0     |
| INR                  |                                    | 0.9 - 1.1   | 1.0   |            | 1.2     |
| Prothrombin time     | s                                  | 10 - 16     | 9.5   |            | 12      |
| APTT                 | s                                  | 24 - 31     | 24    |            | 31      |
| APTT ratio           |                                    | 0.9 - 1.1   | 1.0   |            | 1.2     |
| Urea                 | mmol L <sup>-1</sup>               | 2.5 - 6.7   | 3.2   | 5.0        | 2.7     |
| Creatinine           | mmol L <sup>-1</sup>               | 53 - 97     | 55    | 60         | 57      |
| Sodium               | mmol L <sup>-1</sup>               | 136 - 145   | 137   | 128        | 144     |
| Potassium            | mmol L <sup>-1</sup>               | 3.4 - 5.4   | 4.2   | 4.1        | 3.5     |
| Bilirubin            | µmol L <sup>-1</sup>               | 3 - 21      | 6     | 10         | 19      |
| ALT                  | U L <sup>-1</sup>                  | 10 - 35     | 10    | 7          | 18      |
| Alkaline Phosphatase | U L <sup>-1</sup>                  | 30 - 150    | 58    | 53         | 35      |
| Albumin              | g L <sup>-1</sup>                  | 35 - 50     | 27    | 23         | 20      |
| Calcium              | mmol L <sup>-1</sup>               | 2.10 - 2.60 | 2.18  | 2.0        | 1.69    |
| Calcium ( corrected) | mmol L <sup>-1</sup>               | 2.10 - 2.60 | 2.44  | 2.34       | 2.06    |
| CRP                  | mg L <sup>-1</sup>                 | < 10        | 2.6   | 15.0       | 19.6    |
| Serum Amylase        | U L <sup>-1</sup>                  | 25 - 125    | 58    | 46         |         |
| Glucose              | mmol L <sup>-1</sup>               |             | 7.1   | 8.2        | 10.1    |
| Fibrinogen           | g L <sup>-1</sup>                  | 1.7 - 4.0   |       |            | 2.20    |

## DISCUSSION

The exact cause of development of SAA in pregnancy is not known but is likely to be multifactorial. The hormonal effects of oestrogen and progesterone leading to pathological weakening of vessel wall have been suggested but this hypothesis has been questioned by some authors.<sup>3,4</sup> Failure of elastin production<sup>5</sup>, augmentation of effect of oestrogen and progesterone by the hormone relaxin has also been

proposed as the possible mechanism in pregnancy. Another hypothesis is related to the physiological changes which occur in pregnancy which include increased blood volume and cardiac output. This leads to increased portal flow and splenic arterio venous shunting, which along with vessel wall changes compound the risk of rupture.<sup>3</sup> Aldosterone has also been implicated in causing thinning of arterial vessel walls. Increased intra abdominal pressure, catecholamine release may also contribute to rupture of the aneurysm during pregnancy.<sup>6</sup> Parity was once thought to be an important factor in the development of SAA in pregnancy<sup>7</sup> however some studies contradict this and have found association with nulliparity and low parity. Concomitant non splenic visceral artery aneurysms have been noted in 3% and concomitant non visceral artery aneurysms have been documented in 14%, most frequently involving the renal arteries.<sup>7</sup>

Unfortunately only about 5 % of splenic artery aneurysms present with any symptoms prior to rupture. About 20 - 25% present with the classic 'two stage rupture'. In these patients the aneurysm ruptures giving rise to the initial symptoms. However the blood clot gets contained in the lesser sac because of the clot blocking the Foramen of Winslow or by the omentum. However this ruptures into the peritoneum when the lesser sac ruptures due to tension. This stage is characterised by severe pain and hemodynamic collapse. Sharp epigastric or generalised abdominal pain often gets localised to the left side and can present with left shoulder pain due to diaphragmatic irritation (Kehrs sign). This is accompanied by signs of hypovolemic shock, nausea vomiting and loss of consciousness.

Various atypical presentations of ruptured splenic artery aneurysm in pregnancy have been reported in medical literature. All case reports of ruptured SAA's published so far have report pain to be in the left hypochondrium, epigastric, left flank or generalised abdominal pain radiating to the left shoulder. To the best of our knowledge this is the first reported case of ruptured splenic artery aneurysm of pregnancy presenting as right sided abdominal pain radiating to the right shoulder and arm mimicking a biliary colic. The duration of symptoms for nearly ten days prior to initial presentation is not unheard of in medical literature and patients can have symptoms for weeks.<sup>8</sup> Our patient developed the classic two stage rupture process.

On ultrasound examination, SAA appear as a hypoechoic mass in the left upper part of the abdomen. Duplex

ultrasound examination may show a holosystolic waveform. In case of a ruptured aneurysm the commonest finding might be free fluid in the abdomen. However it might be difficult or at times impossible to see the retroperitoneal structures because of the uterine size or bowel gas. However being a non invasive safe modality in pregnancy, USS remains a modality in use. Our patient had four prior ultrasound scans for antenatal surveillance of IUGR but no abnormal findings were noted. However it is possible that these scans might have missed the findings as they were not aimed at screening for SAA. On CT scans, SAAs appear as well-defined low-density masses with or without calcifications. Intense enhancement within the residual patent lumen following the administration of intravenous contrast medium confirms the diagnosis of an aneurysm. Digital Subtraction Angiography (DSA) remains the 'gold standard' for diagnosis and potential endovascular management of SAA's. In ruptured aneurysms the most suitable option includes an emergency laparotomy with an aim to achieve haemostasis and immediate delivery of the foetus. Haemostasis may be difficult to achieve because of tissue oedema and haematoma. Sometimes division of the stomach, pancreatectomy along with splenectomy may become necessary. Percutaneous transarterial embolisation procedure has been tried with success in a few cases.<sup>9</sup> Splenic artery aneurysms away from the hilum can be excluded with a stent graft. In patients with hilar aneurysms, coil embolisation is a viable option. Embolisation of the entire splenic artery, if selective catheterization of the aneurysm cannot be performed, is an alternative option. Platinum coils, glue, grafts, chemo embolic agents have all been used with varying results. In a recent report of 60 patients who had visceral aneurysms, 16 of 60 (27%) patients were treated with angiographic embolisation. The initial occlusion rate was 81%; recanalisation occurred in 12% of patients.<sup>10</sup> Although Laparoscopic approach has also been tried in these cases it should not be considered in patients who have hemodynamic instability or other signs of rupture.<sup>10</sup> Endovascular treatment is an emerging therapy for splenic artery aneurysms. However endovascular embolisation especially of those affecting the distal splenic artery is prone to develop complications therefore should be avoided. Possibility of splenic infarction, abscess formation, post

splenectomy syndrome and severe pneumococcal sepsis should always be kept in mind after these procedures and appropriate prophylaxis and vaccination should be started.

## SUMMARY

As this case report highlights, ruptured splenic artery aneurysm should be considered as a differential diagnosis in pregnant women presenting with right sided abdominal pain. We wish to stress another significance of this case report, that both mother and the baby had a good outcome considering so few case reports of survival in the medical literature. Our patient despite undergoing massive transfusion developed no coagulopathy and had an uncomplicated recovery. Despite low Apgar score at birth, the baby suffered no adverse outcome. We hope this case report and review improves the awareness of this rather rare but potentially lethal cause of major obstetric haemorrhage in pregnancy.

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