CASE REPORT

Case Report: Successful embolisation of a ruptured splenic artery aneurysm in pregnancy [version 1; peer review: 1 approved with reservations]

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V1 First published: 14 Mar 2019, 8:291
<https://doi.org/10.12688/f1000research.18323.1>
Latest published: 14 Mar 2019, 8:291
<https://doi.org/10.12688/f1000research.18323.1>

Abstract

Background: A case study of a successfully treated rupture of a splenic artery aneurysm during pregnancy, an exceedingly rare condition. The natural history, typical presentation, epidemiology, investigations and available treatments are discussed.

Case: A multigravid 37-year-old presented with acute left upper quadrant pain in her twentieth week of pregnancy. After resuscitation and emergency imaging the patient was diagnosed with a ruptured splenic artery aneurysm. Remaining stable after initial resuscitation allowed for endovascular coiling of her aneurysm.

Discussion: The presentation of ruptured splenic artery aneurysm is rare. While pregnancy is a risk factor, it represents less than 0.1% of pregnancies. However, when it does present it is associated with a high maternal and foetal mortality rate. In this case the stability of the patient allowed for imaging to confirm the diagnosis and the provision of endovascular coiling of the aneurysm. On review of the literature the condition characteristically is diagnosed at laparotomy.

Keywords

Splenic artery aneurysm, pregnancy, endovascular coiling

Open Peer Review

Approval Status ?

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version 1	?
14 Mar 2019	view
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1. Jamil Victor de Oliveira , Centro de Cuidados da Circulação e da Coluna, Sorocaba, Brazil	
Any reports and responses or comments on the article can be found at the end of the article.	

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Author roles: Crawley-Smith T: Writing – Original Draft Preparation, Writing – Review & Editing

Competing interests: No competing interests were disclosed.

Grant information: The author(s) declared that no grants were involved in supporting this work.

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How to cite this article: Crawley-Smith T. **Case Report: Successful embolisation of a ruptured splenic artery aneurysm in pregnancy [version 1; peer review: 1 approved with reservations]** F1000Research 2019, 8:291
<https://doi.org/10.12688/f1000research.18323.1>

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Abbreviations

SAA, Splenic artery aneurysm; G6P3M1T1, Gravid 6 Para 3, 1 miscarriage, 1 termination; BIBA, Brought in by ambulance; LUQ, Left upper quadrant; PV, Per vaginal; SBP, Systolic blood pressure; FAST, Focussed assessment with sonography for trauma; ROTEM, Rotational thromboelastometry; QID, *quarter in die* four times each day

Introduction

Splenic artery aneurysm (SAA) is a rare diagnosis. Its high mortality and typically delayed diagnosis make it a surgical diagnosis not to be missed. While rare during pregnancy, rupture of a splenic artery aneurysm carries a maternal morbidity of 75% and neonatal mortality approaching 95%¹. Within Australia recently there have been two prominent coroners' reports originating from South Australia² and Victoria³. While both coronial inquests recognised the disease's rarity, it was commented that the high morbidity and mortality associated with the condition mandate a high clinical suspicion in the presentation of acute abdomen during pregnancy^{2,3}.

While the most common visceral aneurysm, splenic artery aneurysm is infrequently encountered. Within women of childbearing age it occurs in less than 0.1% of the population⁴, although true prevalence may be unknown, with many being asymptomatic⁵. It has a 4:1 female to male prevalence, and is typically seen at the 6th decade of life⁴⁻⁶. The risk factors include multiparity, angiodyplasia, atherosclerosis, polyarteritis nodosa, cirrhosis, portal hypertension and connective tissue disorders especially α 1 anti-trypsin deficiency⁴⁻⁶. A five-year retrospective study conducted by vascular surgeons in Dallas, Texas, at one of the largest obstetric hospitals within the USA found no diagnoses of SAA for 67,000 births, with only 35 diagnosed cases during that time, none whom were pregnant⁴. 89% of female patients diagnosed had previously been pregnant however, with multiparity an increased risk factor⁴.

The state of pregnancy is a physiological and hormonal risk factor for developing SAA. Tremble and Hill's classic proposal⁷ for the two causes of splenic aneurysm development, being initial weakness of the arterial wall compounded by increased systemic blood pressure, may be applied. During pregnancy, the hormones oestrogen, progesterone and relaxin cause weakness within the vessel wall^{8,9}. Histologically this is described as subendothelial thickening, internal lamina fragmentation, medial myelodysplasia^{8,9} and increased acid glycosaminoglycans within the subintimal and medial layers¹⁰. While the wall is weakened it is vulnerable to the increased pressures associated with the increased cardiac output, circulating volume and portal hypertension associated with the gravid state⁸. Aortic aneurysm, cerebral aneurysm and ovarian aneurysm have also been noted within the literature to be associated with pregnancy and multiparity⁵.

Splenic artery aneurysm may present incidentally during the screening ultrasounds of modern antenatal care, or with rupture^{5,8,9}. The latter is classically described as a presentation of left upper quadrant pain, potentially with Kehr's sign¹¹ (pain radiating to left shoulder tip) with initial instability, a quiescent

phase of bleeding into the lesser sac before further collapse. This double rupture phenomenon is only described in 20–30% of cases⁵ but is important clinically as it may provide a false reassurance to treating staff. The typical size of an aneurysm at rupture is typically above 2.5cm, although ruptures of aneurysms 0.5cm in size have been described. This makes the discovery of the incidental splenic artery aneurysm during antenatal scans problematic, particularly for those under 2cm in size. The diagnosis even at rupture can be a challenge, differentials which can present similarly including pulmonary embolus, amniotic embolism and placental abruption; due to the general instability of the presenting patient imaging may be a guide to treatment but should not interrupt prompt resuscitation. Many descriptions within the literature describe confirmation of the diagnosis at laparotomy and emergency caesarean section.

Patient information

See [Table 1](#) for a timeline of symptoms, diagnosis and intervention.

The patient who presented was a 37-year-old female with significant multigravid obstetric history. G6P3M1T1, she had an inert Mirena *in situ* at the time of her pregnancy and had had two prior presentations to emergency for threatened miscarriage during this pregnancy. She had a previous surgical history of laparoscopic cholecystectomy and Roux-en-Y gastric bypass for obesity, for which she had lost 50kg, current weight being 110kg.

She complained of two days of left upper quadrant (LUQ) pain, no fevers but sweats and vaginal spotting. There were no other reported symptoms of note.

Physical examination

On presentation to emergency with Queensland Ambulance Service she was diaphoretic, in obvious pain localising to the left upper quadrant. She was afebrile, with an initial systolic blood pressure of 80mmHg, which improved to 100-120mmHg with crystalloid resuscitation. Her abdomen was difficult to examine given her habitus, but she had localising peritonism to her LUQ.

Diagnostic assessment

Simultaneous wide bore IV access was obtained while taking venous gas and formal bloods. A cross match was sent. A bedside FAST scan was positive. Obstetric and General surgical review was sought. After two litres of crystalloid resuscitation her blood pressure had stabilised. The initial haemoglobin had come back at 110g/L. A formal ultrasound was ordered within the resus bay. This revealed fluid within Morrison's pouch and a likely splenic artery aneurysm.

Because the patient remained stable, the decision was made to perform a CT angiogram limited to the upper abdomen to limit radiation. The risks and benefits of this were discussed with the patient prior to ordering. This confirmed the diagnosis with a 30x18mm aneurysm arising from the distal splenic artery within the hilum with a small triangular beak anteriorly suspected to be the area of rupture.

Table 1. Timeline of the patient investigation and treatment.

Hour/Day	Event	Investigation/Treatment
Day 0	G6P3M1T1 20/40 pregnant, BIBA with LUQ pain, PV spotting, hypotension SBP 80	Venous blood gas – Hb110g/L Bedside FAST scan – positive for free fluid Foetal heart rate -143bpm
Day 0	Haemodynamically stable after initial 2L crystalloid resuscitation	Obstetric, surgical review Formal ultrasound scan in resus bay- Haemoperitoneum, suspicion splenic aneurysm ROTEM neg E/LFT neg Arterial line.
Day 0	Pt remains haemodynamically stable. Hb stable. Review by Surgical fellow on call; Discussion with Surgical Hepato-Pancreato-Biliary consultant and radiology consultant, laparoscopic management vs angioembolisation	Decision made for CT-Angio to define bleeding; option of embolisation
Day 0		Successfully embolised. Return to surgical ward for observation
Day 1	Review on ward by surgical team and obstetric team	Commenced on full diet. Normal foetal heart rate
Day 2	Ongoing review. Cleared for discharge by obstetric team. Noted to have dysuria	Ultrasound scan to assess flow of splenic artery: no residual flow of artery noted Pt commenced on PO cephalexin 250mg QID
Day 3	Ongoing review. Pt remained haemodynamically stable	Discharged with PO antibiotics, review in clinic in two weeks
Day 14	Reviewed in Obstetric clinic.	Failed to attend surgical Outpatient clinic. Attended Obstetric clinic, ongoing review.

Interventions

Given ongoing haemodynamic stability, there was discussion between the Hepatobiliary consultant, Obstetricians and Intervention Radiology consultant of the risks and benefits of radiographic coiling compared to laparoscopic intervention. The outcome was that an attempt at coiling was made that night with as little radiation and contrast as could be allowed. Where possible single imaging shots were taken rather than continuous runs, with small aliquots of contrast administered in each run. The procedure was successful and the patient was returned to the ward.

Follow up and outcome

After observation for two days, a repeat ultrasound was performed. This showed no residual aneurysm. The patient complained of dysuria at day two and was put on oral cephalexin 250mg QID. She was discharged on day three on oral antibiotics with a plan to attend her obstetric clinic and a surgical clinic at 14 days.

Discussion

While a rare presentation, rupture of a splenic artery aneurysm represents a life-threatening event for the patient and within an obstetric setting, her pregnancy. Prompt diagnosis and resuscitation in this case allowed for further evaluation with further imaging. The fact that the patient remained haemodynamically stable also allowed for the option for interventional radiological coiling. Considerations which were considered by the treating

team were the risk to the foetus from radiation and contrast, combined with the potential delay of surgery in the event of a failed embolisation. The option of laparoscopic evaluation and potential progression to splenectomy was considered. This was to be the next approach in the event of a failed embolisation.

Embolisation of the spleen, while potentially less physiologically stressful than surgical intervention⁸ is not without risk of complication¹. Potential risks included migration of the coil, distal infarction, abscess formation and further aneurysm rupture^{1,5}. The decision to embolise also needs to consider the status of the patient, anatomical location of the lesion and availability of interventional radiology. In the event of instability, prompt surgical intervention, typically through caesarean laparotomy or laparoscopy with ligation of the splenic artery and splenectomy is usually the recommended action.

The mainstay treatment of SAA outside pregnancy is radiological coiling^{5,8}. Introduction of coiling has drastically improved the morbidity and mortality of treatment of visceral aneurysms. Elective treatment should anticipate pregnancy in women of reproductive age with the criteria for intervention typically as a diameter >1cm⁵. Consideration of radiological coiling anticipates the anatomical location of the aneurysm with the goal of spleen preservation, which may not be possible with aneurysms affecting the splenic hilum. The discovery of a splenic artery aneurysm incidentally during pregnancy is worth considering.

Typically, embolisation is recommend above 2cm¹⁰, with intervention ideally occurring in the 2nd trimester, both to minimise harm to the foetus and reduce the risk of rupture, the majority of ruptures being described in the 3rd trimester.

Data availability

All data underlying the results are available as part of the article and no additional source data are required.

Consent

Written informed consent for publication of their clinical details was obtained from the patient.

Grant information

The author(s) declared that no grants were involved in supporting this work.

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Current Peer Review Status: ?

Version 1

Reviewer Report 04 February 2020

<https://doi.org/10.5256/f1000research.20042.r58560>

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The text presents a disease that, although uncommon, is extremely relevant because it affects pregnant women. The text presents a good introduction and clinical description, but it suffers in the part of the intervention, which is poor, without images, without description of the procedure, doses of radiation, contrast, difficulties or technical facilities. In short, there is a lot described in these themes (which after all is the main reason for reading this article). I believe that, if these suggestions are implemented, it will become a rich article, which will be widely read.

Is the background of the case's history and progression described in sufficient detail?

Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

No

Is the case presented with sufficient detail to be useful for other practitioners?

Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Vascular surgeon.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have

significant reservations, as outlined above.

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