CASE REPORT

An unusual diagnosis of splenic rupture

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Accepted 16 September 2014

SUMMARY

A 22-year-old woman presented with a 3-day history of worsening epigastric pain, non-productive cough and vomiting. On examination she was pale and had abdominal tenderness predominant in the right upper quadrant. Abdominal ultrasound excluded the presence of gall stones, but was unable to rule out free fluid in the abdomen. CT demonstrated extensive high-density ascites; however, no source of bleeding could be demonstrated. Clinically the patient's condition deteriorated, and an exploratory laparotomy was performed. In theatre the splenic capsule was found to have detached from the splenic body and emergency splenectomy was performed. Virology serology later demonstrated acute cytomegalovirus (CMV) infection, although tissue microscopy and CMV staining were negative. No other cause of rupture was found. The interesting aspects of this case include the poor correlation between initial presenting symptoms and subsequent diagnosis, the difficulty encountered in making a firm diagnosis and the atypical cause of rupture.

BACKGROUND

Splenic rupture is most typically caused by blunt abdominal trauma. However, numerous other pathologies can result in rupture including infectious, neoplastic, inflammatory or connective tissue disorders. Non-traumatic cases such as these are important to highlight as their presentation may not initially be suggestive of splenic rupture. In these patients, making a firm diagnosis of splenic rupture and investigating the cause of rupture can present challenges. However, the consequences of a missed case are grave; highlighting these issues may prevent future mortality and morbidity.

CASE PRESENTATION

A 22-year-old woman presented to accident and emergency with a 3-day history of dry cough, followed by copious vomiting and severe epigastric pain over the past 1 day. She was otherwise fit and well, had suffered no recent trauma and her only regular medication was a combined oral contraceptive. She had no previous history of reflux symptoms, smoked minimally and had recently been drinking alcohol. On examination the abdomen was maximally tender in the right upper quadrant, and the patient was positive for Murphy's sign. Friends noted that the patient looked paler than usual.

INVESTIGATIONS

A urinary pregnancy test was negative and initial observations were stable. Admission bloods were

non-specific, with a haemoglobin of 12 g/dL, white cells of $13 \times 10^9/L$ and C reactive protein (CRP) of 36 mg/L. D-dimer was significantly raised, at 2120 $\mu g/L$. Owing to the right upper quadrant pain an ultrasound was performed in accident and emergency to rule out cholelithiasis. This confirmed that there were no gallstones present, but was unable to rule out free fluid in the abdomen. The case was discussed with a radiologist, and subsequent CT of the abdomen and pelvis demonstrated extensive high-density ascites with a suspicion that this was blood (22–30 Hounsfield units; [figure 1](#)). There was no free gas, skeletal injury or pelvic abnormality visible, although the ovaries were not seen. The source of bleeding was unclear, although an irregularity at the splenic margin suggested this was possibly the source of haematoma.

TREATMENT

During investigation the patient's clinical status deteriorated. Hypotension and tachycardia persisted despite extensive fluid resuscitation and her haemoglobin dropped to 8.7 g/dL. The decision was made to take her to theatre for exploratory laparotomy. This demonstrated extensive blood and clots within the peritoneum and a ruptured splenic capsule. An emergency splenectomy was performed with minimal further blood loss. Splenic histology subsequently demonstrated that the capsule had largely detached from the surface of the spleen ([figure 2](#)). There was no evidence of rupture to the

Figure 1 CT—coronal section showing massive haematoma with clotting and mass pressure.

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To cite: Roche M, Maloku F, Abdel-Aziz TE. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-204891

Figure 2 Macroscopic histology demonstrating the detachment of the splenic capsule from body and the intact splenic surface.

splenic body and no evidence of focal lesion on serial sectioning. Microscopically, the red and white pulp had normal morphology; there was some focal haemorrhage and exudate on the superficial surface, although this was thought to be postsurgical.

OUTCOME AND FOLLOW-UP

Following surgery the patient was transferred to intensive therapy unit where she subsequently began to develop a temperature. White cells remained raised following surgery, although CRP fell as expected. The patient received 7 days of piptazobactam to little effect. A repeat CT scan performed 6 days postoperatively demonstrated only a small amount of free fluid in the pelvis and partial lower lobe collapse. Virology demonstrated a cytomegalovirus (CMV) viral load of 7000

copies/mL, with CMV DNA found on PCR. CMV IgG and IgM were both positive. Epstein-Barr virus (EBV) and adenovirus viral load were both undetectable, EBV IgM was weakly reactive and IgG positive, consistent with past infection. Microscopic inspection of the splenic tissue did not reveal any CMV inclusions, and tissue staining was negative for CMV and EBV. There was therefore no conclusive cause of splenic rupture diagnosed. There was no indication to treat the CMV infection and fevers resolved during admission. The patient was discharged home 13 days after surgery with appropriate postsplenectomy prophylaxis.

DISCUSSION

Typical symptoms of splenic rupture include left-sided abdominal pain, peritonitic abdomen and hypovolaemic shock. More unusual but well-recognised signs include Kehr's sign (left diaphragmatic irritation resulting in referred left shoulder pain) and Balance's sign (palpable tender mass in the left upper quadrant). This patient did not demonstrate any of these signs, placing her at high risk of misdiagnosis.

We propose that the dry cough the patient presented with was due to blood in the upper abdominal cavity irritating the diaphragm. Subsequent increase in haematoma size led to mass pressure on the stomach, resulting in vomiting and symptoms of reflux. This mass effect is visible on CT imaging. Arguably, it is also possible that pleuritic or gastric infection caused the cough or vomiting and subsequently led to pressure-induced injury to the spleen. However, this theory does not correlate temporally, as the initial symptom was abdominal pain. Further, even minor traumatic injuries typically result in laceration of the splenic surface, rather than the capsular rupture seen here.¹

Causes of splenic rupture can be divided into three categories: traumatic rupture, non-traumatic rupture (infectious, neoplastic, connective tissue disorders, etc) and true spontaneous splenic rupture.²

Splenomegaly is a relatively common complication of CMV infection, affecting around one-third of patients.³ Renzulli *et al*⁴ reviewed 845 cases of atraumatic rupture and found 27% to be due to an infectious cause, although just 1.5% were due to CMV infection (13 cases). In total, 85% of these cases underwent splenectomy.

We performed a literature search using PubMed (search terms 'Splenic Rupture' or 'Splenectomy' and 'Cytomegalovirus' limited to 'English Articles' since '1980'). We found eight previously described cases of splenic rupture found to be secondary

Table 1 Case reports of splenic rupture secondary to CMV infection

	Sex/ age	Co-pathologies	CMV Dx method	Staining/inclusions on histology	Abdominal pathology	Treatment
<i>Medicine</i> (1986) ⁵	M/38	Malaria	Ig	Not documented	Splenic rupture	Splenectomy
<i>Am J Emerg Med</i> (1989) ⁶	F/37	Mitral valve prolapse	Ig	Splenomegaly inclusions	Splenic rupture	Splenectomy
<i>J Infect</i> (1994) ⁷	M/25	Epilepsy	Ig/pathological	CMV inclusions, staining negative	Splenic rupture	Conservative
<i>Am J Kidney Dis</i> (2000) ⁸	M/27	Chronic renal failure/peritoneal dialysis	Ig/PCR	Not documented	Haemoperitoneum	Conservative
<i>Scand J Inf Dis</i> (2001) ⁹	F/43	Tetany, tubal ligation	Ig/PCR	Staining positive	Splenic rupture	Conservative
<i>Presse Med</i> (2007) ¹⁰	F/24	Iron deficiency anaemia	Ig/PCR	Not documented	Splenic rupture	Conservative
<i>Presse Med</i> (2007) ¹⁰	M/29	Migraine, testicular torsion	Ig/PCR	Not documented	Splenic rupture	Conservative
<i>Coll Antropol</i> (2010) ¹¹	M/44	Nil	Ig	Not documented	Splenic rupture	Conservative

CMV, cytomegalovirus; F, female; M, male.

to CMV infection (see table 1).^{5–11} Of these, only two required splenectomy, the others being managed conservatively. Only two cases reported histological findings, with one each being positive for CMV inclusions or staining.

In 1958, Orloff and Peskin published a review of cases that claimed to be truly spontaneous and developed criteria for this diagnosis. These were:

1. On thorough questioning either before or after the operation there should be no history of trauma or unusual effort which conceivably could injure the spleen.
2. There should be no evidence of disease in organs other than the spleen, which is known to affect the spleen adversely.
3. There should be no evidence of perisplenic adhesions or scarring of the spleen, which suggest that it has been traumatised or had ruptured previously.
4. Other than the findings of haemorrhage and rupture, the spleen should be normal on macroscopic as well as microscopic examination.

In 1991, Crate and Payne¹² added a further criterion:

5. Full virological studies of acute phase and convalescent sera should show no significant rise in antibody titres suggesting recent viral infection of types known to be associated with splenic involvement.

This was included as they felt that histological changes resulting from acute infection may be delayed. The case we present in this report does fulfill this fifth criterion; therefore, although we have no direct evidence of CMV infection affecting the spleen, CMV-induced splenic rupture is the most suitable diagnosis.

Management of a patient with splenic rupture is a matter of debate and decisions should be based on the presumed cause, the patient's clinical status and clinician preference. Splenectomy is the classical treatment as it immediately removes the source of haemorrhage and also allows inspection of the abdomen for other sources of bleeding. In our case this was deemed to be the most appropriate course of action as the patient had become haemodynamically unstable and imaging had failed to identify a conclusive source of bleeding. The drawbacks of this approach include high rates of postoperative complication including haemorrhage, sepsis, subphrenic abscess and deep vein thrombosis. Patients are also subsequently at increased risk of serious infection (*Streptococcus pneumoniae*, *Neisseria meningitidis* and *Haemophilus influenza* in particular), needing life-long antibiotic treatment.

The treatment of haemodynamically stable patients is more contentious and can often include conservative management (requiring bed rest and fluid resuscitation as required).¹³ Recently arterial embolisation has been used.¹⁴ These approaches negate the risks of major surgery and also the patient does not subsequently require life-long antibiotic treatment. However, there is no established protocol for following these patients up, no data on the risks of subsequent rupture to a previously injured spleen and no knowledge of the immunological function that a previously ruptured spleen retains.

In summary, we have presented a case of splenic rupture in a previously healthy patient without any of the typical features on history or examination. This case highlights that patients can present with atypical symptoms and that even radiological diagnosis can be difficult if there is no laceration to the splenic body.

Additionally, we highlighted that there are multiple causes for splenic rupture, and these must be explored thoroughly during history and examination. It is important for clinicians to recognise splenic injury quickly and reliably in order to prevent rapid clinical deterioration.

Learning points

- ▶ Rapid and accurate diagnosis of splenic injury is necessary to prevent significant morbidity and mortality.
- ▶ Patients can present with a variety of atypical symptoms, resulting in significant barriers to swift and accurate diagnosis.
- ▶ There are multiple causes for splenic rupture, and it is important to thoroughly investigate these during history and examination in a case of splenic rupture.
- ▶ Management of splenic rupture is contentious, but in the haemodynamically unstable patient emergency splenectomy is always indicated.

Contributors MR drafted and revised the case report as well as performing the literature search and analysis. FM and TE revised the draft paper.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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