Ruptured hepatic artery aneurysm precipitated by gangrenous perforated appendicitis: a case report

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Abstract

Introduction
Hepatic artery aneurysms are among the most common visceral artery aneurysms although still relatively rare. Repair of aneurysms greater than 2 cm in diameter is important due to the high rate of rupture and associated mortality. Here we present a case of a sudden rupture of a hepatic artery aneurysm after presentation with a perforated gangrenous appendicitis. There is increasing evidence that expansion and rupture of abdominal aortic aneurysm is related to degradation of elastin and collagen by matrix metalloproteinases. Elastin degradation leads to rupture. Additionally, matrix metalloproteinases activity has been shown to be upregulated by both sepsis and peritonitis.

Case report
An 80-year-old Caucasian man presented as an emergency with a 7-day history of central abdominal pain that had moved to the right iliac fossa and increased in severity over the last 2 days. His admission chest radiograph was unremarkable. A diagnosis of acute appendicitis was made and the patient was booked at 01:00 for a laparoscopy and appendicectomy on the emergency list the following morning. The cause of abdominal aortic aneurysm (AAA) expansion and rupture is an area of ongoing research. The gradual elucidation of the biological processes underlying aneurysm rupture is changing the view that mechanical forces are solely responsible for aneurysm evolution. This study reports a case of a ruptured hepatic artery aneurysm precipitated by gangrenous perforated appendicitis.

Conclusion
Here we suggest that the inflammation from sepsis and peritonitis lead to the activation or upregulation of matrix metalloproteinases, which, via their collagenase activity precipitated aneurysm rupture.

Introduction
Hepatic artery aneurysms are among the most common types of visceral aneurysms, accounting for 20% of the total. Recognised aetiological factors include atherosclerosis, medial degeneration, portal hypertension, trauma, infection, congenital defects, vasculitis and iatrogenic causes. Epigastric and right upper quadrant pain is the most common presentation, although many are found incidentally on routine imaging. The classic Quincke’s triad of gastrointestinal haemorrhage, biliary colic and obstructive jaundice is seen in less than 30% of presentations.

The spontaneous rupture of hepatic aneurysms into the peritoneal space is associated with a high mortality rate of up to 40%; hence, elective repair is generally recommended in aneurysms greater than 2 cm in size.

The cause of abdominal aortic aneurysm (AAA) expansion and rupture is an area of ongoing research. The gradual elucidation of the biological processes underlying aneurysm rupture is changing the view that mechanical forces are solely responsible for aneurysm evolution. This study reports a case of a ruptured hepatic artery aneurysm precipitated by gangrenous perforated appendicitis.

Case Report
An 80-year-old Caucasian man presented as an emergency with a 7-day history of central abdominal pain that had moved to the right iliac fossa (RIF) and increased in severity over the last 2 days. He was anaemic, nauseated with no diarrhoea or vomiting. His observations were: blood pressure 112/86 mmHg; pulse 90/minute; respiratory rate 18/minute; oxygen saturations 98% on air and temperature 35.9°C. His abdomen was distended and he had signs of localised peritonism in the RIF. His blood tests showed a raised C-reactive protein of 35 mg/L, white cell count of 14.1 × 10^9/L and haemoglobin of 14.2 g/dL. His admission chest radiograph was unremarkable. A diagnosis of acute appendicitis was made and the patient was booked at 01:00 for a laparoscopy and appendicectomy on the emergency list the following morning.

The patient was stable overnight but suddenly deteriorated on the ward at 08:00. He became haemodynamically compromised with a blood pressure of 85/60 mmHg; pulse of 135/minute; saturations of 95% on air and a respiratory rate of 35/minute. An arterial blood gas revealed a lactate of 4.5 mmol/L. A diagnosis of septic shock was made and he was resuscitated with intravenous fluids and antibiotics before immediate transfer to theatre for an emergency laparotomy.

On opening the abdomen, there was a large volume of intra-peritoneal blood but also localised faecal contamination in the RIF due to a necrotic, perforated appendix. The bleeding was localised to the lesser sac in the right upper quadrant. The appendix was gangrenous and perforated. The patient required a total of 11 units of packed red cells. He made a slow but uneventful recovery and was discharged 19 days later.

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upper quadrant. The patient’s abdomen was packed and the appendicectomy completed. As the source of the bleeding was unclear, the patient was transferred for an emergency computed tomographic angiogram without prior closure of his abdomen. This showed a leaking saccular common hepatic artery aneurysm measuring 25–30 mm in diameter. Prompt coil embolisation packing was arranged using a transfemoral approach until no bleeding was evident after 5 minutes of observation. On return to theatre for abdominal closure, haemorrhage was noted after removal of packs surrounding the aneurysm and surgical repair of the aneurysm was required. The patient required a total of 11 units of packed red cells. He made a slow but uneventful recovery and was discharged 19 days later (Figure 1).

**Discussion**

It is recognised that aneurysms can rupture at any size suggesting that the traditional purely mechanical explanation for aneurysm rupture is insufficient. It has been suggested that AAA expansion and rupture is related to increased enzymatic activity at the aneurysm wall. Specifically there is increased proteolysis due to elevated concentration and activity of matrix metalloproteinases (MMPs). MMPs are a group of zinc-dependent proteases that act upon extracellular matrix components including elastin and collagen, which are found in abundance in the arterial wall. The same pathological processes behind degenerative aneurysms have been reported in aneurysms at different sites making it reasonable to assume that research findings from one type of aneurysm can be related to those at other sites.

As an AAA expands its elastin content decreases and, in response to the increase in wall stretch, its collagen concentration increases. Elastin is thought to be responsible for load-bearing at physiological pressures whilst collagen takes over this function at higher pressures. The degradation of elastin leads to aneurysm dilatation whilst the degradation of collagen leads to rupture. Prior to rupture AAAs contain a predominance of collagen types I and III. These end-stage aneurysms are composed of extensive inflammatory infiltration and these inflammatory cells release cytokines that produce and activate proteolytic enzymes such as MMPs. MMP-8 and 9 have both been implicated in AAA rupture. They are both known to have collagenase activity and their elevation is well described in AAA disease. MMP-8 is a potent type 1 collagenase whilst MMP-9 acts on partially degraded fibrillar collagen fragments.

Interestingly for this case, MMPs have been shown to be involved in both peritonitis and sepsis. Serum MMP-8 and 9 levels were shown in one study to be significantly higher in patients with severe sepsis than in healthy controls, suggesting that they may contribute to the host response during sepsis. Additionally, peritoneal mesothelial cells produce MMPs, and levels and activity of MMP-9 are both raised in peritoneal fluid during active peritonitis. It is a source of controversy whether distant injury can specifically increase aortic collagenase activity and more research is needed in this area to support this, although cytokines are well known to mediate systemic effects.

This case suggests that the rupture of pre-existing aneurysms can be precipitated by systemic sepsis or peritonitis. The precise mechanism is unclear but review of the literature suggests this may be due to induction...
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of an inflammatory response that can upregulate collagenase activity at distant sites. This relationship between inflammation and aneurysm rupture has significance for patients with active inflammatory conditions and known aneurysmal disease.

Conclusion

Further research is needed into the rate of rupture of aneurysms in patients with an active inflammatory process as well as the role of MMPs in aneurysmal disease in order that targeted therapies can be produced to down-regulate MMP activity. A number of agents already show promise in this regard. Doxycycline has been shown to be a non-specific MMP-inhibitor capable of stabilising aneurysm size and statins down-regulate the production of MMP-9 in AAAs.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

References