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Spontaneous retroperitoneal haemorrhage from pancreatoduodenal artery (PDA) rupture and associated complications

Edmund Wooi Keat Tan [1,2] Vishal G Shelat,[1,2] Amelia Yuting Monteiro,[3] Jee Keem Low [1,2]

[1]National Healthcare Group, Singapore
[2]Hepatopancreatobiliary Surgery, Tan Tock Seng Hospital, Singapore
[3]Duke-NUS Graduate Medical School, Singapore

Correspondence to
Dr Edmund Wooi Keat Tan; tanwkedmund@gmail.com

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SUMMARY

Spontaneous retroperitoneal haemorrhage (SRH) is rare. It may present with abdominal or back pain with or without haemodynamic instability. Aggressive resuscitation while investigating the cause of bleeding and providing haemostasis are the standard of care. Subsequent close monitoring is necessary to identify early complications.

This study reports three patients who presented to our institution within the last 5 years with SRH from a ruptured pancreaticoduodenal artery (PDA) aneurysm. Each patient had a unique presentation, complications and treatment demonstrating the variability and complexity of SRH. One patient presented with sudden abdominal pain and hypovolaemic shock, underwent angioembolisation and had an eventful recovery. Another patient presented similarly and was treated via angioembolisation but experienced gastric outlet obstruction and obstructive jaundice requiring surgical haematoma evacuation. Another patient had an incidental finding of haemoperitoneum during laparoscopic cholecystectomy that was subsequently diagnosed as SRH resulting from a PDA aneurysm rupture secondary to medial arcuate ligament syndrome.

BACKGROUND

Spontaneous retroperitoneal haemorrhage (SRH) is a rare condition that results from bleeding from retroperitoneal structures. SRH may present with vague symptoms, including abdominal, back or groin pain; blue-grey discolouration of the flank (Grey Turner's sign); decreased haemoglobin or haemodynamic instability. Common causes of SRH include traumatic vascular injury, ruptured abdominal aortic aneurysm, retroperitoneal neoplasms (renal or adrenal) and coagulopathies.[1]

SRH from inferior pancreatoduodenal artery (PDA) aneurysms are rare as PDA aneurysms account for approximately 2% of all splanchnic aneurysms.[2] PDA aneurysms are typically associated with trauma, surgery, endoscopic retrograde cholangiopancreatography of the pancreas, pancreatitis or systemic vasculitis. Previous reports of inferior PDA aneurysms are mostly associated with coeliac axis stenosis or occlusion due to atherosclerosis. CT using the arterial phase can be used to diagnose SRH and localise the site of bleeding so that subsequent therapeutic interventions can be planned.[3]

The treatment of SRH includes the immediate resuscitation and restoration of the circulating volume, urgent radiological interventions and haemostasis. Surgery is reserved for patients in whom haemostasis by interventional radiology modalities fails or those with associated complications such as abdominal compartment syndrome. Following haemostasis, close monitoring for complications such as rebleeding, abdominal compartment syndrome, gastric outlet obstruction (GOO) and biliary obstruction is necessary.

In this study, we present three patients with SRH due to inferior PDA aneurysm rupture. These patients presented to our institution within the last 5 years and demonstrated diverse aetiologies and complexities in clinical management.

CASE PRESENTATION

Patient 1

A woman in her 50s with no relevant medical history presented with acute generalised abdominal pain radiating to both flanks. On examination, she was hypotensive with a blood pressure of 86/46 mm Hg and had significant generalised abdominal tenderness and guarding. No medical or surgical history including prior trauma, pancreatitis or autoimmune diseases were reported.

Patient 2

A woman in her 60s with no relevant medical or surgical history developed sudden onset of severe epigastric pain and tenderness that radiated to her back. On examination, she was haemodynamically stable but had significant epigastric tenderness. No signs of peritonitis were observed, and no abdominal masses were located. The patient reported no history of trauma, pancreatitis, autoimmune disorders or coagulopathies and no use of any traditional or anti-inflammatory medications.

Patient 3

A man in his 50s without significant medical or surgical history presented with right upper abdominal pain and fever. The physical examination revealed tenderness along the epigastrium and right hypochondrium with guarding. The patient denied a history of trauma, pancreatitis and autoimmune disorders. Ultrasound findings led to a diagnosis of acute cholecystitis, and laparoscopic cholecystectomy was scheduled for the following day.

During laparoscopy, significant haemoperitoneum was noted on entry into the peritoneum. The preoperative imaging was reviewed, confirming no

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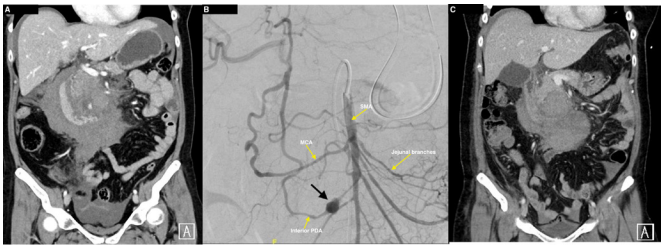

Figure 1 (A) CT coronal section: large retroperitoneal haematoma around the uncinate process of the pancreas. (B) Angiography: aneurysm (black arrow) along inferior pancreatoduodenal artery branch (PDA). MCA, middle colic artery; SMA, superior mesenteric artery. (C) Repeat CT coronal section: retroperitoneal haematoma seen around the second and third part of the duodenum.

preoperative evidence of free fluid in the abdomen. A second hepato-pancreato-biliary surgeon was consulted, and a diagnosis of gallstone-related haemorrhagic pancreatitis was proposed as the hemoperitoneum and bruising were observed around the head of pancreas.

As the patient was haemodynamically stable, the laparoscopic cholecystectomy was completed and a postoperative CT angiogram was scheduled. The cholecystectomy was successful with no complications.

INVESTIGATIONS

Patient 1

Laboratory tests revealed a low haemoglobin (10.6 g/dL), while renal function tests, liver function tests and the serum amylase level were unremarkable. An ultrasound revealed fluid in the retroperitoneum around the right kidney. Urgent CT imaging was obtained, revealing active bleeding in the region of the pancreatic uncinate process with associated fluid in the retroperitoneum (figure 1A).

Patient 2

Emergent CT imaging revealed a large haematoma over the retroperitoneum that extended into the lesser omental sac with no evidence of active bleeding (figure 2A). On day 2 of hospitalisation, the patient reported worsening abdominal pain and had tachycardia (134 beats per minute) and a significant drop in haemoglobin compared with baseline (15.0 to 8.1 g/dL).

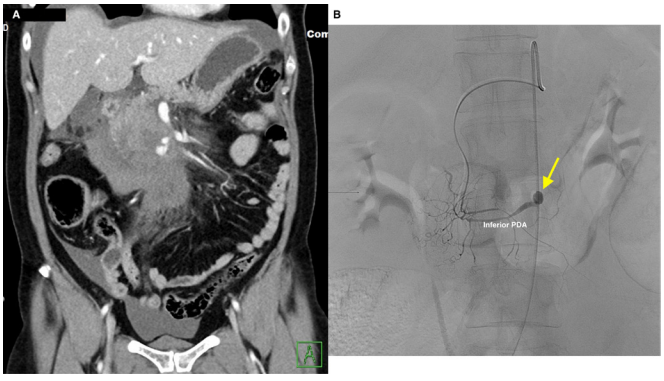

Figure 2 (A) CT coronal section: large retroperitoneal haematoma around the duodenum and pancreas. (B) Angiography: aneurysm (arrow) along the inferior pancreatoduodenal artery (PDA).

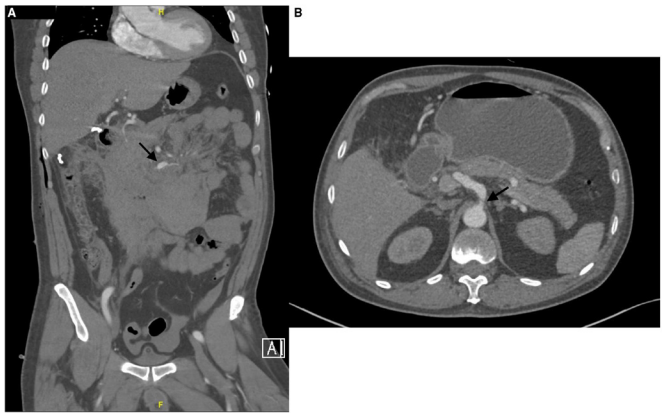

Figure 3 (A) CT coronal section: a massive retroperitoneal haematoma and inferior PDA aneurysm (arrow). (B) CT axial section: a coeliac axis stenosis near the origin with a hypodense linear band-like structure. (arrow). PDA, pancreatoduodenal artery.

Patient 3

Postoperative CT imaging revealed a retroperitoneal haematoma of 17 cm×3.7 cm×9.4 cm with extension into the small bowel mesentery and an inferior PDA aneurysm (figure 3A,B). There was no active contrast extravasation suggesting active bleeding.

The CT images were reconstructed into three-dimensional images (figure 4A). The patient was diagnosed with coeliac stenosis, likely due to medial arcuate ligament syndrome (MALS) based on a hypodense linear band-like structure demonstrated on the CT scan (figure 3B).

TREATMENT

Patient 1

Transcatheter angiography was performed, localising the source of the bleeding as an inferior PDA aneurysm (figure 1B). Embolisation was performed successfully.

Patient 2

An emergency transcatheter angiography was performed. An actively bleeding inferior PDA aneurysm was observed and promptly embolised (figure 2B).

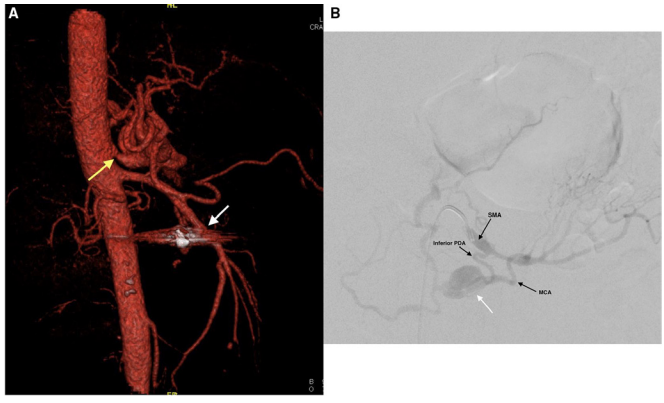

Figure 4 (A): Three-dimensional CT recon (Saggital view) showing coeliac axis stenosis (yellow arrow); inferior PDA aneurysm postangioembolisation (white arrow). (B) Angiography: aneurysm (white arrow) along inferior PDA. MCA, middle colic artery; PDA, pancreatoduodenal artery; SMA, superior mesenteric artery.

Patient 3

Transcatheter angiography was performed, confirming coeliac artery stenosis (figure 4B). Angioembolisation of the inferior PDA aneurysm was successful.

OUTCOME AND FOLLOW-UP

Patient 1

Following angioembolisation, the patient developed worsening early satiety with multiple episodes of bilious vomiting. CT images were obtained on day 4 of the patient's illness, revealing a persistent retroperitoneal haematoma in the region of the uncinate process of the pancreas, encasing the second and third part of the duodenum, resulting in GOO. The patient was managed conservatively with the initiation of total parenteral nutrition and enteral nutrition via a nasojejunal tube.

After 6 days, the patient's symptoms improved, and a gastrografin contrast study confirmed the resolution of GOO. She was discharged and remains symptom-free. A CT scan of the abdomen obtained 6 months postoperatively suggested complete resolution of the retroperitoneal haematoma, and no suspicious masses were observed in the uncinate process. No pathology was observed along the coeliac axis or superior mesenteric vessels. An autoimmune vasculitis screen was negative. The cause of the inferior PDA aneurysm remains unclear.

Patient 2

The patient was unable to tolerate oral feeding following the angioembolisation. A repeat CT scan was performed 3 days after angioembolisation, revealing a large retroperitoneal haematoma involving the second part of the duodenum, resulting in GOO with concomitant biliary obstruction and extrahepatic biliary tree dilatation (figure 5A). Initially, the patient was managed conservatively with parenteral nutrition via a nasojejunal tube. However, the patient's symptoms did not improve after 4 weeks of nutritional support.

A 4-week postoperative CT image revealed persistence of the large retroperitoneal haematoma surrounding the duodenum with worsening biliary dilatation and liver function (figure 5B). The size of the retroperitoneal haematoma was stable. The patient underwent diagnostic laparoscopy and laparoscopic evacuation of the haematoma. Postoperatively, her oral intake and liver function gradually improved.

Two months later, a CT scan suggested complete resolution of the retroperitoneal haematoma. This patient had a similar presentation to patient 1, as both patients had no significant medical history or pathology to suggest a cause of the PDA aneurysm.

Patient 3

The patient developed recurrent episodes of vomiting on postoperative day 5. On postoperative day 6, CT images revealed persistent retroperitoneal haematoma that was unchanged in size, extrinsically compressing the duodenum, resulting in an upper intestinal obstruction. A gastrografin contrast study confirmed delayed transit of the contrast at the second part of the duodenum.

The patient was managed conservatively with parenteral nutrition and a small amount of enteral nutrition. His gastrointestinal function was restored after 5 weeks, and he remains symptom-free at 6 months postoperative.

No autoimmune conditions were found. The patient declined treatment for MALS to prevent recurrences of inferior PDA aneurysms, though he attends regular follow-up visits for surveillance of any inferior PDA aneurysm recurrences.

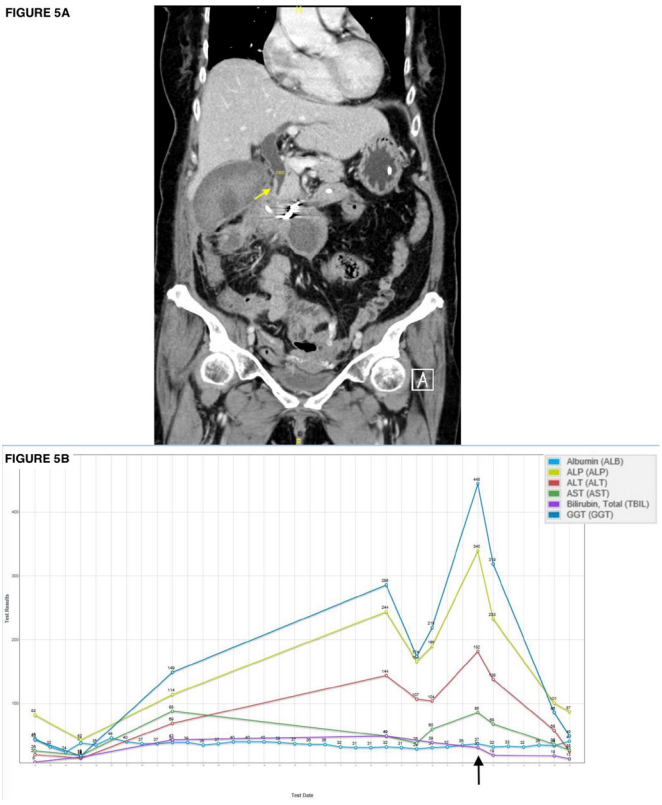

Figure 5 (A) Postangioembolisation CT: retroperitoneal haematoma encasing the second part of the duodenum and distal common bile duct (CBD) with upstream dilatation of the biliary ducts (arrow). (B) Liver function tests trend throughout admission (arrow indicating the time of surgical intervention). ALP, alkaline phosphatase; ALT, alanine transaminase; AST, aspartate aminotransferase; GGT, gamma-glutamyl transferase.

DISCUSSION

In this case series, each patient had SRH of the inferior PDA, though the presentation, complications and management differed. The inferior PDA supplies the pancreas and duodenum, forming an arterial arcade between the areas perfused by the coeliac artery and the superior mesenteric artery (SMA) in the head of the pancreas.

Aneurysms of the inferior PDA are typically associated with atherosclerosis, coeliac axis stenosis, pancreatitis, mycotic or bacterial infections, or trauma. Unlike other aneurysms, the risk of rupture is independent of aneurysmal diameter.[4] PDA aneurysms have been associated with blood flow redistribution due to coeliac trunk stenosis.[4] Previous studies have suggested that haemodynamic changes in aneurysm formation are likely due to increased blood flow in the collateral arteries from the SMA to the coeliac artery and that increased arterial wall shear stress is responsible for aneurysm development, growth and rupture.[5]

The pathogenesis of coeliac trunk stenosis may be intrinsic, such as atherosclerosis or dysplasia, or extrinsic, such as median arcuate ligament compression, which is observed in 10%–24% of patients with coeliac stenosis.[6] Divergent blood due to stenosis of the coeliac trunk results in a high flow rate and turbulent blood flow in smaller branches of the SMA,

leading to the development of erosion and the formation of aneurysms.[7]

The gold standard imaging of patients with PDA aneurysms is CT with arterial phase. The resulting images provide essential information that is required to localise the bleeding site and plan subsequent therapeutic interventions.[3] Immediate resuscitation and haemostasis are the standard of treatment. Surgery should be considered when haemostasis cannot be achieved using interventional radiology modalities or when complications, such as abdominal compartment syndrome, are present.

No treatment guidelines have been established for the management of PDA aneurysms. Most experts agree that these aneurysms must be treated once detected; however, very few are found early enough to prevent rupture. Approximately 7%–15% of PDA aneurysms are associated with gastrointestinal haemorrhage,[8] and the bleeding is mainly into the retroperitoneal space. When a PDA aneurysm ruptures, the mortality rate is up to 50%.[5]

The most concerning complication after transcatheter embolisation is a subsequent rupture with bleeding into the abdominal cavity, which occurs in 5%–20% of patients.[9] Therefore, these patients must be monitored carefully.

Several previous case reports suggest various treatment strategies for the obliteration of coeliac trunk stenosis, resolution of any associated pathologies and maintenance of adequate blood flow to territories of the coeliac trunk. The surgical options include ligation and resection and aneurysmorrhaphy of the PDA aneurysm. However, these treatments are associated with high mortality rates and technical difficulties, especially after rupture, and are not commonly performed.[10] Therefore, the ideal treatment of a PDA aneurysm is intravascular aneurysm embolisation for both ruptured and unruptured subtypes on detection. Intravascular treatment, such as stenting for coeliac artery stenosis, may help restore blood flow. However, in patients in whom the cause of the stenosis is extraluminal, such as MALS, the standard therapy may be MAL incision surgery.[11]

After the initial diagnosis of SRH and treatment, clinicians should also monitor the patients carefully for any complications, especially when the initial bleeding event resulted in significant blood loss or large haematomas were observed on initial imaging. All three patients in this case series developed intestinal obstruction several days after treatment. One patient also developed concomitant biliary obstruction. These obstructions are primarily due to the compressive effects of the massive retroperitoneal haematoma. The time between the onset of GOO symptoms and the initial presentation of SRH ranges from 10 to 14 days. The mechanism of obstruction is associated with two primary anatomic factors: the fixed retroperitoneal position of the second to the fourth part of the duodenum and the location of the inferior PDA, which is closely related to the duodenum and biliary structures.

In patients with persistent symptoms suggestive of intestinal obstruction, further evaluation of the upper gastrointestinal tract is necessary to exclude other common causes of gastroduodenal obstruction. Additional investigations, including CT scans, gastrointestinal contrast studies or endoscopy, are warranted. When patients experience obstruction, conservative and surgical approaches must be considered. The preferred methods of conservative management include nasogastric tube suctioning and supportive therapy with intravenous fluids, enteral feeding via nasojejunal tubes (bypassing the obstructed portions of the duodenum) and intravenous parenteral nutrition supplements. The duration of conservative treatment is dependent on several factors, including complications such as sepsis from an infected haematoma, concomitant biliary obstruction and abdominal compartment syndrome. The location and feasibility of evacuating the haematoma via percutaneous or surgical means also impact the duration of conservative treatment.[12] When conservative treatment fails, more invasive options should be considered.[13 14]

Learning points

- Spontaneous retroperitoneal haemorrhage from inferior pancreatoduodenal artery (PDA) aneurysms is rare, and the mainstay of treatment is immediate resuscitation, emergency imaging to localise the source of bleeding and haemostasis.
- Massive blood loss or large haematomas observed on initial imaging are associated with a higher risk of complications from extrinsic compression of surrounding structures and may be deadly.
- After successful haemostasis, patients should be monitored closely, and clinicians should retain a high index of suspicion for any associated complications.
- Inferior PDA aneurysms are associated with trauma, surgery, endoscopic retrograde cholangiopancreatography, pancreatitis, infection, systemic vasculitis and coeliac artery stenosis or occlusion. Therefore, any underlying predisposing causes should be evaluated and treated accordingly to reduce the risks of recurrence.

Contributors EWKT: data curation, formal analysis, investigation, project administration, resources, visualisation, writing–original draft. VGS: data collection, supervision, validation, writing–review and editing. AYM: formal analysis, investigations, writing–review and editing. JKL: data collection, conceptualisation, methodology, supervision, writing–review and editing.

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Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

ORCID iDs
Edmund Wooi Keat Tan http://orcid.org/0000-0002-3425-7432
Jee Keem Low http://orcid.org/0000-0002-1090-9336

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