CASE REPORT

Inferior phrenic artery pseudoaneurysm complicating drug-induced acute pancreatitis

Jean F Salem,¹ Ali Haydar,² Ali Hallal¹

¹Department of Surgery, American University of Beirut Medical Center, Beirut, Lebanon

²Department of Diagnostic Radiology, American University of Beirut Medical Center, Beirut, Lebanon

Correspondence to

Dr Jean F Salem, js56@aub.edu.lb

SUMMARY

Inferior phrenic artery (IPA) pseudoaneurysm is an extremely rare complication of chronic pancreatitis with only three cases reported in the literature so far. It is a serious condition that can be life-threatening if not diagnosed promptly. Recent advances in endovascular interventions made angiography with embolisation the modality of choice for diagnosis and treatment. We presented the first report of a case of ruptured IPA pseudoaneurysm complicating a drug-induced acute pancreatitis that was successfully treated by transcatheter arterial embolisation. Despite its rarity, rupture of pseudoaneurysm due to drug-induced pancreatitis should be suspected and included in the differential diagnosis when associated with haemodynamic instability.

BACKGROUND

We presented the first report of a case of ruptured inferior phrenic artery (IPA) pseudoaneurysm complicating a drug-induced acute pancreatitis that was successfully treated by transcatheter arterial embolisation.

It is a life-threatening condition that requires prompt diagnosis and treatment to prevent fatal outcome.

CASE PRESENTATION

A 19-year-old female patient known to have T-cell lymphoma, on chemotherapy (L-asparaginase) and low-molecular weight heparin for subclavian veins thrombosis, presented with diffuse abdominal pain, nausea and vomiting of several days' duration.

There was no fever, haematemesis, melaena or jaundice.

Physical examination revealed severe right lower quadrant tenderness. Laboratory studies revealed a white cell count of 11 900/dL, haemoglobin 12.5 g/dL, platelets 165 000/dL, serum bilirubin 0.8, alanine aminotransferase 167 IU/L, amylase 98 IU/L and triglyceride 489 mg/dL. Contrast-enhanced CT examination of the abdomen revealed an enhancing thickened collection posterior to the uncinate process of the pancreas extending into the perirenal fascia and in the paracolic gutter. In addition, there was also a subscapular liver collection around segment VI. The overall radiological findings together with the patient's clinical presentation and the history of L-asparaginase made drug-induced pancreatitis our provisional diagnosis. Two days later the patient developed worsening abdominal pain, which was associated with haemodynamic instability and a drop in the haemoglobin to 6.6 g/dL. A CT angiography was performed which revealed active bleeding into a fluid collection lying inferior to the pancreas (figure 1). A selective digital subtraction angiography showed a pseudoaneurysm of a branch of the IPA with active contrast extravasation (figure 2).

Embolisation of the bleeding vessel using coils was successfully achieved (figure 3). The patient's haemodynamic parameters and clinical condition stabilised after embolisation and resuscitation with blood product. She subsequently developed gastric outlet obstruction due to a large haematoma in the retroperitoneum which was compressing the duodenum. She was managed non-operatively with nasogastric tube insertion and parenteral nutrition.

Figure 1 CT angiography (A: axial view, B: coronal view) revealing active bleeding (vascular blush) into a collection lying inferior to the pancreas (arrows).

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Figure 2 Selective digital subtraction angiography showing pseudoaneurysm of a branch of the inferior phrenic artery with active contrast extravasation (arrow).

Ten days later, the patient started tolerating diet and was then discharged home in stable condition.

DIFFERENTIAL DIAGNOSIS

- ▶ L-asparaginase-induced pancreatitis complicated by massive bleeding from a ruptured IPA pseudoaneurysm.
- ▶ Spontaneous bleeding from an IPA pseudoaneurysm.

OUTCOME AND FOLLOW-UP

On follow-up, 5 months later, the paediatric team reintroduced L-asparaginase as it was the preferred treatment of her disease.

L-asparaginase was used again as there was no hard evidence that it had induced pancreatitis. Unfortunately, the patient presented again a few days after the reintroduction of L-asparaginase in her treatment with severe pancreatitis documented biochemically with an amylase of 434 IU/L and lipase of 1206 U/L and radiologically with a CT scan showing necrotising pancreatitis (figure 4).

DISCUSSION

Pseudoaneurysm complicating chronic pancreatitis is a serious condition with an incidence of 10%.¹ It is very rare in acute

Figure 3 Selective digital subtraction angiography after coiling of the bleeding branch (arrow) showing complete occlusion of the pseudoaneurysm.

Figure 4 CT scan showing acute necrotising pancreatitis.

pancreatitis. The condition is thought to result from proteolytic digestion of the wall structures of vessels. Its rupture may be life-threatening with a mortality rate ranging from 12% to 57% in treated cases and may be as high as 90–100% in untreated ones.² The splenic artery is involved in 60–65% of cases because of its contiguity with the pancreas, followed in decreasing order of frequency by the gastroduodenal artery (20–25%), the pancreaticoduodenal artery (10–15%), the hepatic (5–10%) and the left gastric arteries (2–5%).³

IPA pseudoaneurysm is an extremely rare condition with only three cases reported in the literature.^{3, 4} Our case describes the first IPA pseudoaneurysm complicating a drug-induced (L-asparaginase) acute pancreatitis.

Angiography is considered the gold standard for diagnosis of pseudoaneurysm and is an important treatment modality. Contrast-enhanced CT angiography is becoming a first-line non-invasive diagnostic modality for localising the site of pseudoaneurysm and for planning further management. In our case, the diagnosis of pancreatitis and pseudoaneurysm was suggested based on a constellation of indirect signs: history of treatment with L-asparaginase (15% risk of pancreatitis), physical examination and finding of peripancreatic inflammation and collection around the head of the pancreas. It was later confirmed by the development of frank pancreatitis after the readministration of L-asparaginase. The amylase level was normal due to the delayed presentation of the patient. The contrast-enhanced CT

angiography showed bleeding within the collection but failed to determine the bleeding vessel accurately. Angiography was then performed and established the definitive diagnosis of active bleeding from the IPA. The bleeding vessel was embolised with coils and haemostasis was successfully achieved. Surgery is an alternative therapeutic option but with higher morbidity and mortality rates.³ The success rate of angiographic embolisation ranges from 79% to 100% and thus becomes the modality of choice in the treatment of pseudoaneurysms.³

The most serious and life-threatening complication of pseudoaneurysms is rupture with a mortality rate approaching 100% if left untreated.² Fortunately, and despite the delayed diagnosis, our patient remained stable over several days because the IPA ruptured in the retroperitoneum and the haematoma were contained. The success of the embolisation in our case was confirmed by follow-up CT scans that revealed resolution of the haematoma and absence of contrast extravasation.

Learning points

- ▶ Inferior phrenic artery pseudoaneurysm is an extremely rare complication of chronic pancreatitis.
- ▶ This first described case shows that it can also complicate acute pancreatitis.
- ▶ It is a life-threatening condition. Urgent diagnosis is essential.
- ▶ CT angiography is the first-line diagnostic modality.
- ▶ Angiography with embolisation is the modality of choice for treatment especially in haemodynamically unstable and high-risk patients.

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