

Case Report

Rare complications of a low lying median arcuate coeliac ligament

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ABSTRACT

Pancreaticoduodenal artery aneurysm is a rare complication of coeliac artery stenosis secondary to a low lying median arcuate coeliac ligament. This article reports the case of a 69-year old man who presented with left arm and leg weakness, clinically in keeping with right hemisphere stroke. Initial CT brain scan was within normal limits. The patient did not receive thrombolysis as he was outside the time window. 3 hours later the patient experienced sudden onset epigastric



Fig 1. Sagittal CT reconstruction showing the coeliac axis (C) and superior mesenteric artery (S); The typical shape of the coeliac axis as it passes under the median arcuate ligament is marked by the arrow.

pain and acute shock. CT aorta abdominal was diagnostic of a ruptured inferior pancreaticoduodenal artery aneurysm. Repeat CT brain the following day showed subacute infarction within the right frontal lobe. Embolisation of the aneurysm was successfully performed. It is well documented that ischaemic stroke can cause acute hypertension. This acute hypertension probably contributed to the rupture of the pancreaticoduodenal artery aneurysm. The patient was well on discharge and remains well 2 months on.

CASE REPORT

A 69 year old man was admitted with left sided arm and leg weakness of 5 hours duration. He had a past medical history including ischaemic heart disease, CABG, hypertension hypercholesterolaemia and asthma. He was taking aspirin 75mg once daily. He lived with his wife and was independent for all activities of daily living.

Examination showed no cranial nerve abnormality. Medical Research Council (MRC) grading system for power scored 5/5 in the right upper and lower limbs and 4/5 in the left upper and lower limbs. Achilles tendon reflexes and deep tendon reflexes were increased on the left with a left sided upgoing plantar reflex. He was hypertensive at 206/125. The patient was not examined for the presence of an abdominal bruit.

The patient's initial full blood counts and biochemical parameters were within normal limits. Computed tomogram of the brain was also normal. The clinical picture was consistent with a diagnosis of right hemisphere lacunar stroke. As the patient was outside the thrombolysis time window, he was treated with 300mg aspirin.

3 hours after presentation, the patient developed sudden onset epigastric and back pain. He appeared cold and clammy. His blood pressure fell to 74/48 and radial pulses were not palpable. CT abdominal angiogram showed a ruptured inferior pancreaticoduodenal artery aneurysm. Coeliac axis

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stenosis with a configuration typical of a low lying median arcuate ligament was also identified. Serum amylase was normal and there was no clinical evidence of current or previous pancreatitis. The ruptured aneurysm was not embolised at this time as the patient was stable and it was felt a multidisciplinary decision should be made of the best course of action.

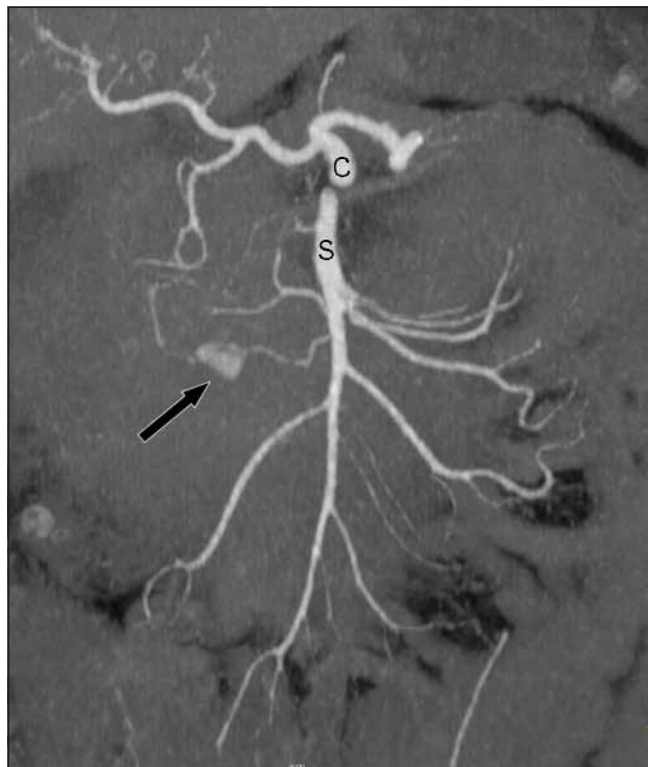


Fig2. Coronal CT reconstruction showing the coeliac axis (C) and superior mesenteric artery (S); the aneurysm (arrow) arises from the inferior pancreaticoduodenal artery which is a branch of the superior mesenteric artery.

The following day repeat CT brain revealed subacute infarction within the right frontal lobe, in the right anterior cerebral artery territory. After liaison between an interventional radiologist, surgeon and stroke physician, 300mg aspirin OD was continued. Echocardiogram showed mild left ventricular hypertrophy and mild dilatation of the ascending aorta. Carotid Doppler ultrasound revealed only mild (<30%) stenosis in both internal carotid arteries.

Repeat CT aortogram 3 days later showed persistence of the inferior pancreaticoduodenal artery aneurysm, again with evidence of recent rupture and haemorrhage. The patient proceeded to mesenteric catheter angiography which revealed a pancreaticoduodenal aneurysm, coeliac artery stenosis and the development of large pancreaticoduodenal territory collateral vessels supplying the coeliac vessels from the superior mesenteric artery. The pancreaticoduodenal aneurysm was successfully embolised

The patient was discharged with the support of the stroke early supported discharge rehabilitation team. At 2 month review, he has a very mild residual left sided weakness but

is independent of all activities of daily living. He has not experienced any further abdominal pain.

DISCUSSION

Compression of the coeliac axis by a low lying median arcuate ligament in asymptomatic patients has long been recognised and is a common finding on modern sagittal CT reconstructions.¹ Findings on CT include focal narrowing of the coeliac axis with poststenotic dilatation and increased collaterals from the superior mesenteric artery.

In most people, the median arcuate ligament passes superior to the origin of the coeliac artery, near the first lumbar vertebra. However 10-24% of the population may have an abnormally low ligament.² Complications of this anatomical variation are described and include dilatation of the pancreaticoduodenal collateral pathways and subsequent formation of a visceral aneurysm. The pancreaticoduodenal artery is a branch of the superior mesenteric artery (SMA). The superior pancreaticoduodenal is a branch of the gastroduodenal artery (supplied by the coeliac artery); the inferior pancreaticoduodenal artery is a branch of the SMA. These two form an arcade that allows collateral supply from the SMA to support the coeliac territory, which is undersupplied from coeliac artery due to the stenosis caused by the low lying median arcuate ligament. It is postulated that the increased flow through this collateral pathway results in the hypertrophy of the vessels and true aneurysm formation. Visceral aneurysms are a rare but potentially lethal form of vascular disease, with an incidence of 0.01% to 0.2% in routine autopsies.³ Pancreaticoduodenal artery aneurysms are estimated as 2% of all visceral aneurysms.⁴ The risk of rupture of these aneurysms is uncertain as the number of cases is too small to determine a trend.⁵ Rupture is however well described and carries a 29% mortality rate.⁶ Contemporary management is usually by emergency coil embolization.³

A less well understood complication is median arcuate ligament syndrome or Dunbar syndrome. It is the triad of abdominal pain, weight loss and abdominal bruit in patients whose only positive imaging finding is coeliac artery stenosis. The pathophysiology of this condition is contentious as mesenteric ischaemia is commonly held to only occur when at least two of the three mesenteric vessels are compromised. Our patient did not display the classical symptoms of this syndrome.

Hypertension is commonly observed in the immediate post-stroke period.⁷ As described by Brainin and Heiss, 82% of patients had systolic blood pressures above 140 mmHg during the first 48 hours, 28% having a systolic blood pressure above 180 mmHg.⁸ Cerebral perfusion becomes dependent upon systemic arterial BP following stroke due to impairment of cerebral autoregulation. Hypertension may sustain cerebral perfusion to the ischaemic penumbra. Very high blood pressure has been associated with poor post-stroke outcomes. Poor outcomes are said to be due to a number of factors including vascular complications and early stroke recurrence.

It is likely that the hypertension following our patient's stroke caused rupture of the preexisting pancreaticoduodenal artery aneurysm given the rarity of visceral artery aneurysm rupture. Administration of thrombolysis as a part of stroke management may have resulted in an adverse outcome.

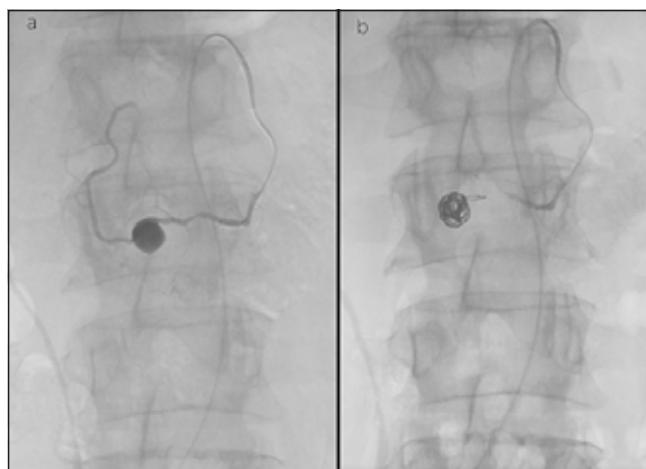


Fig 3. (a) catheter angiogram of the inferior pancreaticoduodenal artery demonstrates the aneurysm. (b) the aneurysm has been filled with embolization coils and there is no contrast flow into the aneurysm.

There are no previously documented cases of stroke and rupture of a pancreaticoduodenal artery aneurysm presenting together. Our case highlights the difficult management issues including thrombolysis contraindications and management of these aneurysms.

LEARNING POINTS

1. Low lying median arcuate anatomy associated with coeliac stenosis is a common incidental finding on modern imaging, rarely associated with pancreaticoduodenal artery aneurysm.

2. Incidental diagnosis of an asymptomatic pancreaticoduodenal artery aneurysm warrants prompt evaluation and consideration of treatment.
3. Symptomatic rupture of a pancreaticoduodenal artery aneurysm is exceptionally rare.
4. Hypertension is commonly observed in the immediate post-stroke period. This has been associated with poorer outcomes.

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^a20 mg once daily for the prevention of stroke and systemic embolism in adult patients with non-valvular atrial fibrillation with ≥ 1 risk factor. AF, atrial fibrillation; PE, pulmonary embolism.