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A case of Pancreatic duct aneurysm causing massive gastrointestinal (GI) bleeding treated by Whipple procedure

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Abstract

Gastrointestinal (GI) bleeding is a common clinical encounter referring to a symptom of disease rather than a disease itself. Most GI bleeding sites are usually located using endoscopy, colonoscopy or angiography. However, there are some situations, in which the bleeding sites remain very difficult to locate. We report a case where a male patient presented with sudden onset of fresh blood in stool and a history of recurrent melena. Gastroscopy revealed superficial ulcers and blood clots at the duodenum and the duodenal-jejunum junction respectively. Computed Tomography (CT) and Magnetic resonance imaging (MRI) showed pancreatic duct dilation. Digital Subtraction Angiography (DSA) could not identify any bleeding sites and the patient was discharged after bleeding had stopped. Post-discharge, there was massive haematochezia and CT of the pancreatic head identified an aneurysm around the pancreatic duct. After ruling out all

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potential sites for the massive haemorrhage, a pancreatic duct aneurysm was considered as the bleeding source and emergency laparotomy was performed. Intraoperatively, the pancreatic parenchyma was found to be oedematous and very prone to bleeding; therefore, the surgeon opted to proceed with a pancreaticoduodenectomy (Whipple procedure). Following the usual approaches for a Whipple procedure, the surgery went on well and the bleeding was stopped. This is a unique entity of how a visceral-arterial aneurysm communicating with the main pancreatic duct was managed.

Keywords: Gastrointestinal bleeding, computed tomography, digital subtraction angiography, pancreatic duct aneurysm, pancreaticoduodenectomy (Whipple procedure).

INTRODUCTION:

An aneurysm occurs due to pathological changes within the wall of an artery leading to the widening of its lumen[1]. Rupture of these aneurysms along the GI tract will cause symptoms of GI bleeding and may present with the acute clinical symptoms of shock[2]. Most aneurysms can be detected by CT scans and angiography[3]. Some are however difficult to locate and the treatment becomes problematic. True aneurysms of the pancreaticoduodenal arcades are very rare.

CASE REPORT:

A 68 years old male patient presented with sudden onset of blood in stool for one day and was experiencing melena for one month. The patient had a history of hypertension, cerebral infarction, diabetes, and an unknown source of gastrointestinal bleeding but no history of hematemesis. He was not on anticoagulant therapy. He had an acute episode of pancreatitis, which resolved after treatment at a local hospital 5 years ago. On admission, physical examination was unremarkable with no signs of fever and jaundice. Routine blood test showed: Hb 59 g/L, RBC $2.36x10^{12}$ /L, Hct 19.00%, WBC $8.61x10^{9}$ /L, PLT $162x10^{9}$ /L. Routine urine test: urine glucose level (+++) Blood

biochemistry: total bilirubin 6.30umol/L, direct bilirubin 3.00umol/L, blood glucose 15.5umol/L, CA19-9 33.69 U/L.

Emergency gastroscopy on the first day of admission revealed a small superficial solitary ulcer measuring 3mm x 8mm on the anterior duodenal wall surrounded by congestive and edematous mucosa and no other abnormalities were detected in the descending part of the duodenum. The patient was given blood transfusion and fluid infusion until there was sufficient circulatory dilatation and, with continuous infusion of PPI and somatostatin to stop the bleeding. On the second day, MRI revealed an atrophied pancreas with dilatation of the pancreatic duct at both the body and the tail (Fig. 1).

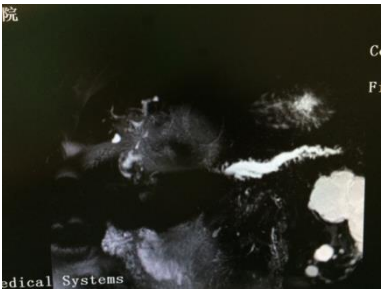

Figure 1. MRI showing dilatation of the pancreatic duct in the body and tail + atrophy of the pancreas.

On the 3rd day of hospitalization, the patient had another episode of massive haematochezia. Duodenoscopy and colonoscopy were done to detect the bleeding source but no abnormalities were found in the stomach, the duodenal bulb, or the descending part of the duodenum. Although there were some traces of 'old blood' in the duodenal-jejunum junction, no obvious bleeding lesions were found. Colonoscopy could not be completed as the colon was filled with blood clots. DSA was performed and branches of the celiac trunk were embolized suspecting possible bleeding sites. No further bleeding sources were found from the superior and inferior mesenteric arteries (Fig. 2,3,4).

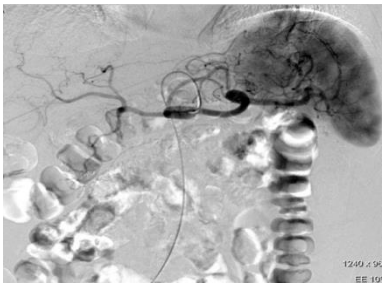

Figure 2. Celiac arteriography + embolization.

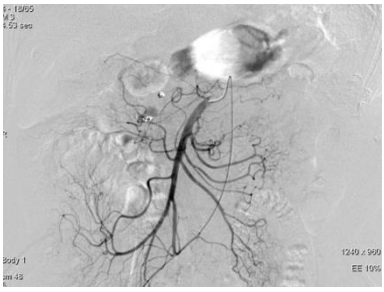

Figure 3. Superior mesentery artery arteriography.

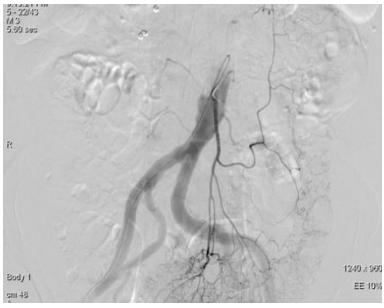

Figure 4. Inferior Mesentery arteriography.

On the fourth day, a CT scan showed pancreatic duct stones and an aneurysm of size 1.0x1.0cm at the head of the pancreas (Fig. 5,6). Post-embolization, bleeding had stopped and the patient was discharged after 5 days. Six days after discharge, the patient had another episode of massive haematochezia and, developed dizziness, fatigue, and palpitation.

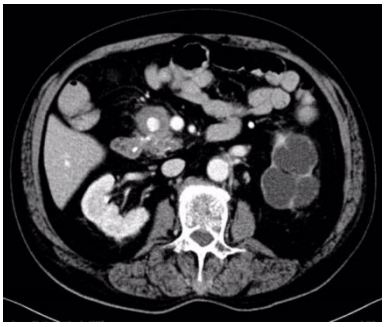

Figure 5. CT showing an aneurysm at the head of pancreas.

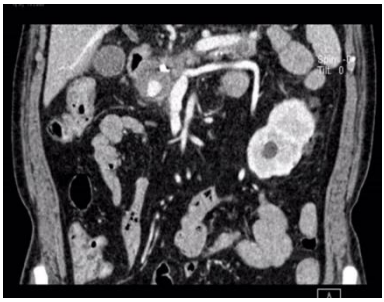

Figure 6. CT showing an aneurysm at the head of pancreas on 4th day of admission.

On re-admission, laboratory tests showed: RBC routine test: Hb 55 g/L, RBC $2.26x10^9$ /L, Hct 17.90%.

The patient was given blood transfusion, IV fluids, and coagulating factors. DSA was performed again and arteriography of the celiac artery together with both the superior and inferior mesenteric arteries did not show any active bleeding site. On the next day, the patient lost another 800 ml of blood in stool, with a significant drop in blood pressure. As DSA did not reveal any ongoing site of haemorrhage, a possible diagnosis of bleeding from the pancreatic aneurysm was suspected. Embolization did not deem useful in this situation because the aneurysm had many important and different branches from the pancreaticoduodenal arcade. An emergency laparotomy was performed without further delay. Chronic inflammation of the pancreas made the parenchyma oedematous and very prone to bleeding, and as such, dissecting and separating the pancreatic head was unsafe. It was difficult to accurately locate the

visceral arterial aneurysm due to easy bleeding and consequently the aneurysmal body was not found. In these circumstances, the surgeon opted to proceed with a pancreaticoduodenectomy. Following the usual approaches for a Whipple procedure, the surgery went on well and the bleeding was stopped. Macroscopically a haemangioma in the head of the pancreas, accompanied by haemorrhage and necrosis was identified (Fig. 7).

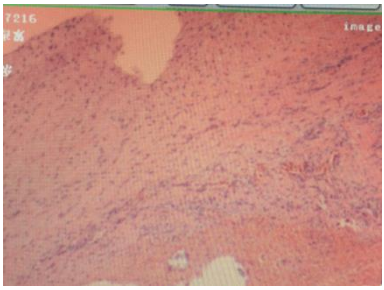

Figure 7. Hemangioma of the head of pancreas accompanied by hemorrhage and necrosis.

Pathological results confirmed that the mass was a ruptured aneurysm (Fig. 8). Postoperatively, the patient had a 10-month follow-up without any signs of recurrent bleeding.

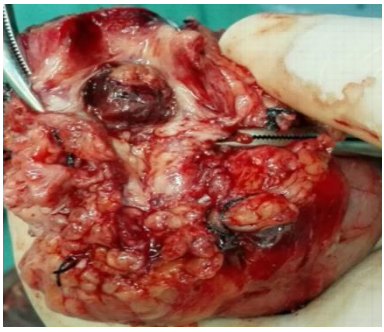

Figure 8. An aneurysm of about 1.0x1.0cm was found in the pancreatic head and communicating with the pancreatic duct.

DISCUSSION:

Pancreatic duct aneurysm could happen because of increased flow in the small fragile pancreaticoduodenal arteries in the presence of

celiac, common hepatic related or superior mesenteric arteries stenosis/occlusion[2]. Initially, the vessels enlarge to adapt to increased flow and the vessel wall is weakened leading to a true aneurysm formation due to the continuous increased flow and high intra-arterial pressure[4]. It has been reported that hypertension is an important factor in aneurysm development of pancreaticoduodenal arteries while chronic pancreatitis is a possible cause of formation of pancreatic pseudo-aneurysm[5, 6]. Large aneurysms may be visible on ultrasound examination with the use of colour flow imaging and diagnosed on aortography but an aneurysm and its parent arterial supply are best assessed with contrast-enhanced CT. The gold standard for diagnosing small aneurysms is conventional catheter angiography and selective catheter injections through the celiac, gastroduodenal, superior mesenteric, and inferior pancreaticoduodenal arteries [3].

In our case chronic pancreatitis, hypertension and diabetes could be the contributing factors to the pancreatic duct aneurysm. Our initial diagnosis was GI bleed but DSA could not identify the source of haemorrhage. Factors that cause bleeding in the oesophagus, stomach and duodenum were not considered, as gastroscopy was normal. Although the first gastroscopy found a small solitary ulcer, the second gastroscopy confirmed that there were no other bleeding ulcers or vascular malformations. Duodenoscopy found some old blood near the duodenal end indicating that this case was an Upper GI bleeding. Colorectal bleeding was excluded due to the presence of dark blood clots and no active bleeding sites were found. CT scan did not show any small intestinal tumor or anomalies and no arterio-venous malformations were found during the superior and inferior mesenteric arteriography. As such, small intestinal bleeding was also eliminated. Moreover, although bleeding was recurrent and similar to the characteristics of periodic bleeding of the biliary tract, the patient had no jaundice, the serum total bilirubin and direct bilirubin were normal, and the gallbladder was not enlarged on imaging investigations. Haemobilia was therefore not considered.

After having interpreted and disregarded all other possible causes of bleeding we began assessing the other features of the bleeding factors in this patient, which would conform to the characteristics of a haemorrhagic aneurysm. The bleeding was fierce

and rapid indicating a possibility of a bleeding aneurysm. Dilatation of the tail end of the pancreatic duct seen on MRI suggested that the aneurysm most probably was located in the parenchyma of the pancreatic head. The blood from the ruptured aneurysm would flow in the pancreatic duct itself before following its course to the small intestine through the duodenal papilla. The bleeding was periodic probably because of the minimum pressure required for the ampulla to open and the blood to pass through.

Although we confirmed the presence of a pancreatic aneurysm, it seemed difficult to precisely have access to it. Studies have suggested that therapeutical approaches can be either surgical resection/ligation or transcatheter embolization[7-9]. In some cases, pancreaticoduodenectomy can be performed depending on the remoteness of the aneurysm location[10].

The possibility of successful embolization and haemostasis was small as an arcade of arteries including both the superior and inferior pancreaticoduodenal arteries was supplying the aneurysm. Vessels within the collateral web of arteries may have enlarged and lead to the formation of the aneurysm at the pancreatic duct[10, 11]. While we had chosen to proceed with surgery, we found out that the pancreatic parenchyma was oedematous with severe adhesion, very prone to bleeding and it was difficult to isolate the bleeding source. Taking into consideration the images of CT and MRI scans, Whipple procedure was deemed to be the best choice to stop the bleeding from the pancreatic duct. Despite not being able to find the haemorrhagic point, Whipple procedure proved to be efficient in stopping the bleeding. We consider this case to be rare, and suggest that pancreatic duct aneurysm to be included as a differential diagnosis for an unknown source of GI bleed and could also be used as a reference in the future.

Declarations:

Ethics approval and consent: This manuscript has been reviewed and approved by the ethics committee of Nanjing First hospital affiliated to Nanjing Medical University.

Financial Support: None to declare.

Conflicts of Interest: The authors have no conflicts of interest to declare.

Acknowledgement: We thank the patient for giving his consent for the publication of this report.

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