SCIREA Journal of Clinical Medicine

http://www.scirea.org/journal/CM

December 23, 2016

Volume 1, Issue 2, December 2016

Pseudoaneurysm complicating pancreatitis: what is the best treatment? Case presentation and review of the literature.

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ABSTRACT.

Pseudoaneurysms secondary to pancreatitis, acute or chronic, are a rare complication (reported incidence in a range from 1.3% to 10%), potentially lethal. An early detection and intervention, either surgical or by minimally invasive interventional procedure, is vital for the patient. Multidetector Computed Tomography is the gold standard for the diagnosis.

Endovascular therapy with coil embolization is the first-line option for treating pseudoaneurysms subsequent to pancreatitis acute or chronic.

We discuss transcatheter coil embolization treatment and subsequent resolution of a giant pseudoaneurysm resulting from an abnormal pancreatic branch of hepatic artery, with an aberrant origin directly from aorta, caused by an alcoholic chronic pancreatitis in a 35-years-old man with gastrointestinal bleeding and severe anemia.

KEYWORDS: Gastrointestinal Bleeding; Visceral Pseudoaneurysms; Chronic pancreatitis; Coil Embolization; Vascular Complications; Hepatic Artery.

INTRODUCTION.

Pseudoaneurysm is an apparent common vascular complication of pancreatitis with a reported incidence in a range from 1.3% to 10%; it can cause lethal haemorrhage and the mortality in untreated patients can be as high as 90% [1]. For this reason an early detection and intervention, either surgical or by minimally invasive interventional procedure, is vital for the patient.

The splenic artery (SA) is the most commonly affected vessel in up to 50% cases, while pseudoaneurysms arising from the hepatic artery (HA) are very uncommon (2% of cases) [2].

Multidetector computed tomography (MDCT) has become the initial study of choice and characterization. On the basis of MDCT information, conventional angiography can then be performed to confirm and treat pseudoaneurysm [3].

We discuss trans-catheter coil embolization treatment and subsequent resolution of a giant pseudoaneurysm resulting from an abnormal pancreatic branch of HA, with an aberrant origin directly from aorta (type 9 – Michels Classification) [4], caused by an alcoholic chronic pancreatitis in a 35-years-old man with gastrointestinal bleeding and severe anemia. are discussed.

CASE REPORT.

A 35-years-old male with a history of chronic and significant alcohol intake for the past 10 years with a precedent episode of acute pancreatitis two years before, presented in the

emergency department for a severe abdominal pain radiating to the back, hematemesis and melena. The patient reported a severe weakness for the past four months. The patient was anemic (Hb 7.0 g/dL) with an elevated heart rate (130/min) and his blood pressure was 90/60.

Suspecting an upper gastrointestinal bleed the patient underwent to endoscopy unit to perform an upper gastrointestinal endoscopy that was unhelpful. It was decided for an urgent Computed Tomography Angiography (CTA) (LightSpeed VCT 64 Slice General Electric, USA) that demonstrated an atrophic pancreas with multiple calcifications of the pancreatic head and body and in the arterial phase a giant pseudoaneurysm originating from an abnormal pancreatic branch of HA, with an aberrant origin directly from aorta. The total pseudoaneurysm dimension was 76 mm x 58 mm x 55 mm, including the thrombosed part. The only perfuse part measured 50 mm x 35 mm x 38 mm ***(Fig. 1).***

Due to the high risk of rupture the patient was referred to emergency angiography study (Integris, Philips, Eindhoven, The Netherlands) for embolic treatment of the pseudoaneurysm.

Intravascular access was obtained through a right common femoral artery puncture using the modified Seldinger technique, and a 5 Fr femoral arterial sheath was placed.

Diagnostic hepatic angiography was obtained by selective cannulation of HA with a curved F4 Cobra catheter in digital subtraction angiography (DSA)-technique. A giant pseudoaneurysm with a very small neck was identified very close to HA origin from aorta; his parent vessel was an abnormal pancreatic branch deriving from it ***(Fig. 2a)***.

A F2.7 microcatheter (Progreat, Terumo, USA) was advanced into the pseudoaneurysm to identify the outflow vessels. Coil embolization of the only outflow vessel that formed collateral branch with superior mesenteric artery, was performed with two 0.018 inches micro-tornado® pushable steel coils (Tornado® Embolization Microcoil™, Cook incorporated, Bloomington, IN, proximal end diameter 5 mm × distal end diameter 2 mm). The size of micro-tornado coil was selected to match the diameter of the segment to embolize.

Consecutive angiography control was performed in order to check coil placement at the affected site and to demonstrate complete occlusion of the vessel.

Subsequently, the F2.7 microcatheter pulled back into the pseudoaneurysm neck where 3 micro-tornado® coil (Tornado® Embolization Microcoil™, Cook incorporated, Bloomington, IN, proximal end diameter 6 mm × distal end diameter 2 mm) were deployed to close the direct arterial flow. For the high risk of rupture the pseudoaneurysm was not filled with coils.

At the final angiographic control there was immediate technical success with no further inflow into the pseudoaneurysm sac and a normal hepatic artery flow was seen ***(Fig. 2b)***.

A contrast-enhanced follow-up CT scan four days after verified a complete occlusion of the pseudoaneurysm, with thrombosis of the pseudoaneurysmal sac. The patient's ensuing hospital stay was uneventful and he could be discharged after 6 days without any signs of bleeding ***(Fig. 3)***.

DISCUSSION.

Pancreatitis may cause a spectrum of venous and arterial vascular complications, ranging from asymptomatic venous thrombosis to catastrophic variceal haemorrhage and from incidentally discovered pseudoaneurysms that remain stable over years of follow-up to acute life-threatening rupture [5].

Pseudoaneurysms, as vascular complication of pancreatitis, can occur in both acute or chronic ones; however, they are more common in chronic pancreatitis and are often associated with pseudocysts. Pseudoaneurysms are the most dangerous vascular complications of pancreatitis for its high haemorrhage probability [1-2]. Indeed, Bergert et al. reported severe bleeding complications in 36 of 541 patients with chronic pancreatitis, a prevalence of 6.7%; the most common cause of major haemorrhage was pseudoaneurysm present in 25 patients (69.4%) [6].

A pseudoaneurysm is defined as an encapsulated hematoma in communication with the lumen of the ruptured vessel [7], where the external wall consist of adventitia, perivascular tissue, fibrosis, or clot [8].

The exact pathogenesis of pseudoaneurysm formation is still unclear, but, to date, three pathogenic mechanisms are being discussed: (1) severe inflammation and enzymatic autodigestion of a pancreatic or peripancreatic artery may cause a disruption of the artery; (2) an established pseudocyst eroding a visceral artery, thereby converting the pseudocyst into a large pseudoaneurysm, and (3) a pseudocyst may erode the bowel wall with bleeding from the mucosal surface itself [9-10].

In our case, in the history patient there was a previous acute pancreatitis two years before, with the formation of a pseudocystic lesion. It is thought that the pancreatic juice within this pseudocyst caused in time enzymatic degradation of the wall of adjacent hepatic artery, with weakness and rupture leading to pseudoaneurysm formation.

The SA is the most commonly affected vessel in up to 50% cases, given its anatomic location. Gastro-duodenal artery (GDA) pseudoaneurysms are seen in up to 20% of cases, and pancreatico-duodenal (PDA) arteries are involved in up to 10% of cases. Pseudoaneurysms arising from the superior mesenteric artery and HA are very uncommon, with a reported incidence of 3% and 2% respectively. Aortic involvement is quite rare, with a reported incidence of 0.5% [2-5]. A single left phrenic artery pseudoaneurysm was noted in only one case report [10].

Pseudoaneurysms subsequent to pancreatitis are associated with a high mortality, that can reach 90% if not treated; then, an early diagnosis is essential [6-7].

Currently, the best method of assessing pancreatitis vascular complications is MDCT, providing a clear road map for endovascular therapy with a high sensitivity of 95% and a specificity of 90%. CT can be performed quickly and has a high reported accuracy in detecting arterial complications. MDCT accurately localizes the pseudoaneurysm and its arterial supply, identifies the difficult angle of origin or an anomalous origin of artery allowing us to draw up a clear strategy for the approach and which embolization technique to use, before the procedure [12].

We recommend a biphasic protocol with a portal venous phase, as small pseudoaneurysms and those with narrow necks may become significantly more conspicuous during portal venous phase imaging. This additional phase is also useful in the evaluation of venous thrombosis that can be missed if CTA alone is performed.

DSA allows imaging in real time, helping to correctly identify the artery of origin and assess the collateral circulation. DSA is more sensible (almost 100%), especially when there are very small and tiny pseudoaneurysms that may occasionally be missed on CTA, providing at the same time a chance of treatment. However DSA underestimates the size of a pseudoaneurysm with the peripheral thrombus visualizing only the patent lumen whereas CT imaging shows the lumen and the peripheral thrombus and thus the actual size of pseudoaneurysm [13-14].

Hyare et al. retrospectively compared 29 studies in 25 patients with haemorrhagic complications subsequent to pancreatitis in which CTA was performed within 24 h preceding DSA. The sensitivity and specificity of CTA for the detection of major arterial bleeding on a background of pancreatitis were calculated as 0.947 and 0.900, respectively, demonstrating technical accuracy for the detection of major arterial haemorrhage in inflammatory pancreatic disease and concluding that CTA should be consider as the first investigation in the

management of these patients [3].

The management of pancreatic pseudoaneurysms depends on factor such as hemodynamic stability, coagulation status, and source of bleeding. There are many treatment options for pancreatitis-associated arterial pseudoaneurysms: traditional surgical treatment included celiotomy, ligation of the celiac trunk, and partial pancreatectomy; interventional radiology included transcatheter arterial embolization with the use of coils, covered stents, detachable ballons, gel foam or particles or percutaneous/endoscopic thrombin injection [9-12].

Because of the low overall incidence of pseudoaneurysms and the need to achieve rapid haemostasis, randomized control trials of therapeutic options are not possible.

Historically, pseudoaneurysms were surgically repaired, but with modern minimally invasive interventional techniques, surgery is no longer typical first-line therapy [15].

Balachandra et al. demonstrated the presence of a significantly greater mortality rate in patients undergoing surgery as first intervention compared with those undergoing transcatheter arterial embolization, which is a powerful tool for the control of haemorrhage avoiding the need for urgent, high-risk surgery [16].

Moreover, Kriwanek et al. have reported an interesting finding in their study in which bleeding complications were more frequently encountered in patients who underwent early surgical treatment than in those who underwent delayed interventions. They suggested that techniques such as gauze packing and drainage tube insertion performed during necrosectomy and debridement may cause iatrogenic injury to the fragile vessels, therefore increasing the risk of bleeding [17].

Surgically repaired pseudoaneurysms carry their own mortality rates, with wide ranges depending on the case series, between 10% and 50% [18-19].

To date endovascular therapy is the first-line option for treating visceral pseudoaneurysms. The reported success rate of endovascular therapy is high, ranging from 79-100%, with recurrent bleeding rates ranging from 18-37%. The overall mortality related to angiographic failure or a complication was reported to be 14% in patients with chronic pancreatitis, but it depends principally by the experience of the operator [11].

The commonest technique used to isolate the pseudoaneurysm is coils embolization, as in our case, and the implantation of covered stents if larger and life-important arteries are affected in order to exclude the site of bleeding or pseudoaneurysm sac and preserve flow in the parent

vessel. Technically, metallic coil embolization of expendable arteries (that is arteries with an extensive collateral circulation) is preferable distal and proximal to the site of arterial extravasation (the so-called isolation technique), thereby preventing backflow from collateral circulation [10-20].

It is not necessary to fill the aneurysm sac, as long as the efferent and afferent arteries are occluded and the potential for reperfusion is minimized. Packing the sac with coils increases the risk of rupture, is an unnecessary expense, and is time-consuming. In addition, the insertion of the coils into the sac makes follow-up imaging to assess reperfusion very difficult.

While coils are the most widely used embolic material, there have recently been an increasing number of reports describing the use of N-Butyl cyanoacrylate glue (NBCA), especially when feeder distal vessel cannot be accessed [21-22].

NBCA can be used individually or in combination with other embolic materials (generally ethiodized oil - Lipiodol), because it is not radiopaque. The liquid nature of this embolic material allows it to conform according to the anatomy of the pathologic vessel, and it can be delivered distally into narrow or tortuous vessel that would otherwise be difficult, if not impossible, to reach with a microcatheter [23].

If necessary, these materials can be combined each other; the use of coil and embolic material together is so called "sandwich" technique.

Rebleeding is a potential problem with endovascular management. Kalva et al. demonstrated rebleeding rates following embolization at 24 hours and 30 days to be equal to 4% and 17% respectively in a population of 39 patients [24], value very similar to prior reports [25].

Higher rebleeding rates were observed in patients with large pseudoaneurysms related to pancreatic pseudocysts [26].

Complications associated with angiographic embolization, such as intra-procedural rupture of the pseudoaneurysm and delayed reconstitution of arterial flow, result in the failure of treatment. However, these complications are rare and are mostly unrelated to angiographic technique [8].

Minor complications from embolization include focal splenic infarction (only observed among patients who underwent embolization of the SA) with a rate of 15-20; major complications include splenic infarction with possible abscess formation, coil migration, intestinal necrosis and vascular dissection with a rate not up to 5% [23-24].

The percutaneous injection of thrombin into the pseudoaneurysm under US or fluoroscopy guidance can be used for pseudoaneurysms not easily accessible via a trans-catheter technique and in which the feeding vessel cannot be identified. This approach, since involves puncture through the wall of the pseudoaneurysm which may be especially risky when the wall is friable, is preferred in chronic pancreatitis because pseudoaneurysm wall are more thicker compared with pseudoaneurysm in acute ones [27-28].

In the last years numerous case reports about endoscopic US-guided treatment are appearing in the current literature. Numerous authors report injecting glue, alcohol, or thrombin into accessible pseudoaneurysms, predominantly involving the SA. Their procedures have been met with success and stability without rebleeding or requiring surgery [29-30-31-32].

CONCLUSION

Between vascular complication related-pancreatitis pseudoaneurysms are the most common cause of major haemorrhage. Radiologists have to know what is the best modality to diagnose and to treat them.

MDCT is a sensitive and accurate technique for the detection of major arterial haemorrhage in inflammatory pancreatic disease and should be considered as the first investigation in diagnosis and for planning intervention.

Trans-arterial embolization is effective in the management of pancreatitis-related pseudoaneurysms and has good outcomes. Coils embolization is the treatment of choice. The emergency surgical treatment, for its high mortality rates, should be limited to cases where angiography is not available or is failed.

AUTHOR #1 declare no conflict of interest.

AUTHOR #2 declare no conflict of interest.

AUTHOR #3 declare no conflict of interest.

AUTHOR #4 declare no conflict of interest.

AUTHOR #5 declare no conflict of interest.

Informed consent was obtained from the patient included in the study.

"All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards"

FIGURE LEGENDS:

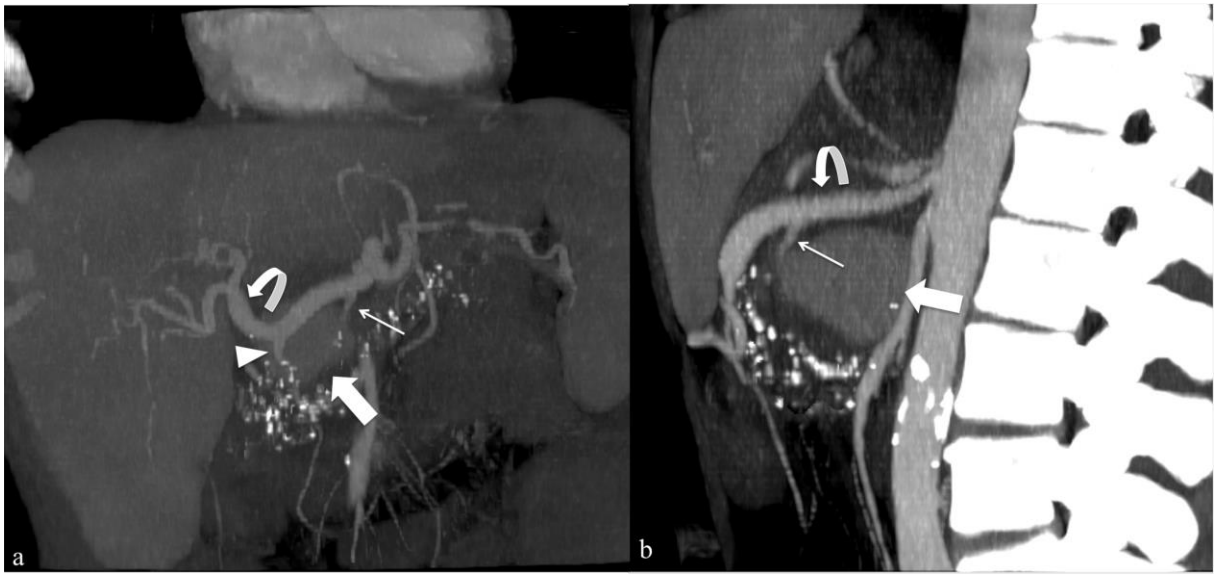

Fig. 1: a-b) Sagittal and coronal contrast enhanced CT (arterial phase) MIP of the upper abdomen shows a giant pseudoaneurysm (arrow) 76 mm x 58 mm in size originating from an abnormal pancreatic branch (thin arrow) of HA (curved arrow). Diffuse and multiple calcifications of the pancreas body and tail suggest the presence of the underlying chronic pancreatitis. Pancreatico-duodenal arcade appear hypertrofic (arrowhead).

Fig. 2: a) Selective HA angiography with a microcatheter (Progreat, Terumo, USA) in DSA-technique (manual injection of 10 ml Imeron 300, Altana, Germany) reveals the pseudoaneurysm and its proximal parent vessel (thin arrow), originating very close to HA outlet directly from aorta; b) Superselective angiogram (manual injection of 10 ml Imeron 300, Altana, Germany) after apposition of micro-tornado® coils (thin arrow) (Tornado® Embolization Microcoil™, Cook incorporated, Bloomington, IN) reveals correct coils placement and complete occlusion of the vessel with a persistent complete patency of the HA and its branchs (arrowheads) and arcade pancreaticoduodenal (arrow).

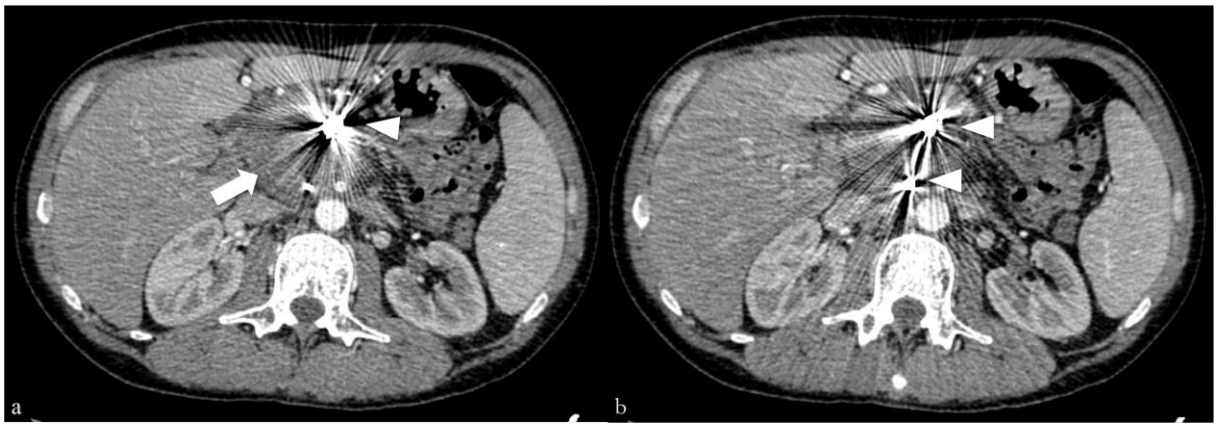

Fig. 3: a-b) CT control after coil embolization of the giant pseudoaneurysm (portal/vein phase) (a upper slice, b lower slice) 2 weeks after embolization. No perfusion of the pseudoaneurysm was found in the arterial, portalvein or delayed phase (arrow). The exam is limitated by coil metallic artifact (arrowheads).

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