

Percutaneous thrombin embolization of a pancreaticoduodenal artery pseudoaneurysm after failing of the endovascular treatment

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Core tip: We present a rare case of pseudoaneurysm of the pancreaticoduodenal artery (PDA) in a patient with no risk factors but with an occlusive lesion of the celiac axis. To the best of our knowledge this is the first reported case of PDA pseudoaneurysm successfully treated in emergency by single transabdominal ultrasonography-guided injection of thrombin after failed attempts of percutaneous catheterization of the feeding vessel of the pseudoaneurysm.

Abstract

Pancreatico-duodenal artery (PDA) pseudoaneurysms are rare vascular conditions with high mortality rates after rupture and they are frequently secondary to pancreatitis, surgery, trauma or infection. Due to the high risk of rupture and bleeding, it is mandatory to treat all pseudoaneurysms, regardless of their size or symptomatology. First option of treatment is open surgical repair, but it has high mortality rate, especially in hemodynamically unstable patients. In the recent years, percutaneous ultrasonography (US)- or computed tomography-guided thrombin injection was proposed as an alternative method for treating visceral aneurysms and pseudoaneurysms, but few reports described this therapy in case of peri-pancreatic pseudoaneurysms. We present a rare case of pseudoaneurysm of the PDA in a patient with no previous history of pancreatitis nor major surgery but with an occlusive lesion of the celiac axis. To the best of our knowledge this is the first reported case of PDA pseudoaneurysm successfully treated in emergency by single transabdominal US-guided injection of thrombin after failed attempts of percutaneous catheterization of the feeding vessel of the pseudoaneurysm.

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INTRODUCTION

Pancreatico-duodenal artery (PDA) pseudoaneurysms are potentially life-threatening but rare vascular conditions, accounting for 2% of splanchnic artery aneurysms and with mortality rates after rupture of 15%-50%^[1-4].

They are mostly due to pancreatitis, surgery, trauma, infection, iatrogenic lesions, vasculitis and atherosclerosis^[5], but also to the presence of a celiac axis or common hepatic artery stenosis/occlusion, known as Sutton-Kadir syndrome^[6,7]. These lesions may be identified by means of different imaging techniques [ultrasonography (US), computed tomography (CT), magnetic resonance

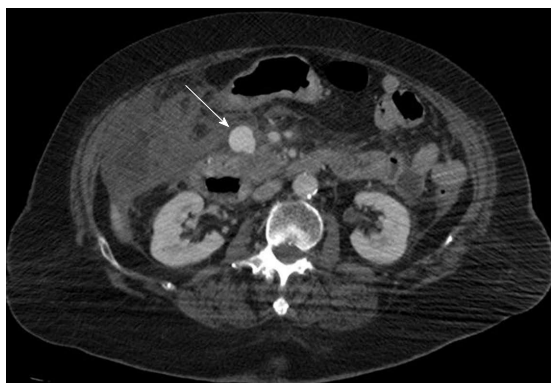


Figure 1 Axial contrast-enhanced computed tomography showing pancreatico-duodenal artery pseudoaneurysm (arrow) before the treatment.

imaging (MRI) and digital subtraction angiography (DSA)].

Due to the high risk of rupture and bleeding and the absence of predictive factors, it is recommended to treat all pseudoaneurysms, regardless of their size or symptomatology^[8]. Open surgical repair, consisting of resection, ligation, exclusion, bypass or endo/organ resection, has high-mortality rate and is applied in hemodynamically unstable patients or as secondary approach after failed transcatheter embolization^[9].

In the recent years, with the development of materials and techniques in the field of interventional radiology, percutaneous endovascular management of such a pseudoaneurysm is becoming the first-line treatment, with low morbidity and mortality^[10,11].

Percutaneous US- or CT-guided injection is an alternative method for treating visceral aneurysms and pseudoaneurysms^[12-23], but few reports described this therapy in case of superior mesenteric artery (SMA) or PDA pseudoaneurysms^[9,24-28].

To the best of our knowledge, this is the first reported case of a PDA pseudoaneurysm percutaneously treated by direct thrombin injection by US guidance after failing of endovascular treatment.

CASE REPORT

An 82-year-old woman was in a peripheral hospital two days before for cerebral haemorrhage and was admitted in emergency to our central hospital with the suspicion of acute bleeding. During the hospital stay routine laboratory investigation revealed severe anaemia with haemoglobin drop to 6.0 g/dL and contrast-enhanced thoraco-abdominal CT scan was performed on a 64-slice multi-detector Siemens scanner. It identified an oval, rounded mass within the pancreatic head with slow centrifugal contrast-enhancement, presumed to be a bleeding pseudoaneurysm with a haematoma and a small haemoperitoneum around the spleen (Figure 1). The feeding artery to the pseudoaneurysm was identified as a PDA, given the area of distribution and the origin from SMA. The patient's surgical history was unremarkable and did not have

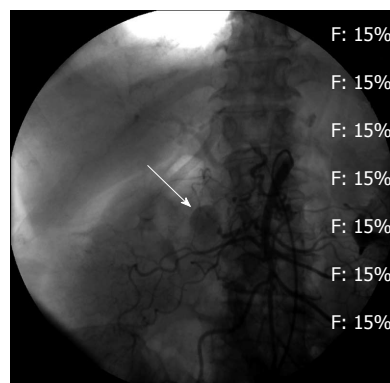


Figure 2 Selective superior mesenteric artery catheterization showing pancreatico-duodenal artery pseudoaneurysm (arrow) but the feeding vessel could not be identified.

history of chronic pancreatitis.

At the admission the patient underwent to endotracheal intubation but was hemodynamically stable. Due to her age and clinical co-morbidities, open surgery was not the best option and an attempt with endovascular procedure was decided.

Angiography was performed using Siemens equipment (Axiom Artis U, Siemens AG, Forchheim, Germany). Under local anaesthesia (lidocaine 2%), using the Seldinger technique, a selective SMA catheterization (Cobra Glidecath, 5F, 65-cm-long, Terumo, Tokyo, Japan) with a hydrophilic guidewire (0.035", 150-cm-long, Terumo) confirmed the pseudoaneurysm of 2-cm on diameter of the PDA area (Figure 2). Both the examination of the SMA made in different projections, including the lateral view and superselective catheterization (Progreat, 2.7F, 130-cm-long, Terumo, Tokyo, Japan) of at least 4 proximal branches did not show the pseudoaneurysm so that the feeding vessel could not be identified.

Selective catheterization (Radifocus guidewire, 0.035", 150-cm-long, and Cobra Glidecath, 5F, 65-cm-long, Terumo) of the celiac artery (CA) was performed, but the gastroduodenal artery (GDA) was not depicted, because of a close stenosis of the take-off of CA from the abdominal aorta.

Since we were not able to identify and catheterize the feeding vessel, and considering the patient's high-risk clinical condition, percutaneous embolization with thrombin injection was suggested. The procedure was performed after local anaesthesia (lidocaine 2%) and a Chiba needle (Ecojekt 18G, 20-cm-long, HS Hospital Service, Aprilia, Latina, Italy) was inserted into the pseudoaneurysmal sac through the anterior abdominal wall under US guidance (MyLab, Esaote, Genova, Italy) and 2 mL (1000 IU) of human thrombin (Tisseel, Baxter AG, Rome, Italy) were injected. During the injection, the anechoic area of pseudoaneurysm turned into hypoechoic area and the colour signals disappeared because of immediate thrombosis was achieved on the US control (Figure 3).

A final SMA selective control DSA performed at the end of the procedure confirmed complete exclusion of

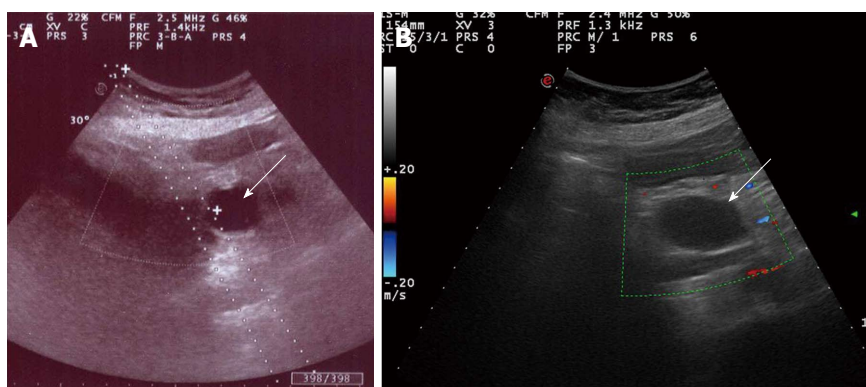


Figure 3 Two-dimensional B-mode ultrasonography (A) and color-Doppler ultrasonography (B) showing pancreatico-duodenal artery pseudoaneurysm (arrows) before and after percutaneous thrombin injection, respectively.



Figure 4 Axial contrast-enhanced computed tomography showing pancreatico-duodenal artery pseudoaneurysm (arrow) 1 wk after the treatment.

the pseudoaneurysm with no endoleak and preserved perfusion of the SMA.

Finally, haemostasis of the puncture site at the right common femoral artery (CFA) was obtained using a vascular closure device (Angioseal 6F, Saint Jude, MN, United States).

Thrombotic or immunologic complications of thrombin were not observed. A 1-wk follow-up abdominal CT scan showed a completely thrombosed PDA pseudoaneurysm with no endoleak or recurrence of bleeding, and preserved flow through the SMA branches (Figure 4). Patient's condition was stable and the blood test normalized. Doppler ultrasound examinations at 2 wk and 3 mo confirmed thrombosis of the pseudoaneurysm, without any echographic signs of blood flow within.

Patient was discharged from the intensive care unit and she is now under careful clinic and imaging follow-up.

DISCUSSION

Natural history of the visceral false aneurysms is potentially the rupture and a lethal abdominal haemorrhage, with a rupture rate of 50% and a mortality rate up to 80%^[29].

Causes of SMA aneurysm and pseudoaneurysm formation include chronic pancreatitis, atherosclerosis, trau-

ma, surgery, biopsy, infection, collagen vascular disease, medial necrosis, arthritis and dissection.

Symptoms depend on the location and include abdominal pain, melena, body loss, internal haemorrhage and hypovolemic shock due to pseudoaneurysm rupture.

Our patient did not have history of chronic pancreatitis and was suspected to have an internal haemorrhage because of onset of severe anaemia and lacking a worsening of the brain haemorrhage.

Imaging techniques for the diagnosis of pseudoaneurysms include US, CT, MRI and DSA^[4]; this shows precise location, morphology, size of the sac, feeding collateral vessels and efferent arteries if present, and enables immediate endovascular treatment.

PDA or SMA pseudoaneurysms require immediate treatment due to a high mortality, up to 90%, in the case of not treatment^[18] and it includes open surgical repair and non surgical procedures.

Surgical treatment, traditionally a first-line therapy option^[30], is invasive and is associated with high risk of infection, haemorrhage and long rehabilitation time. Deep retroperitoneal and retroduodenal position of the vessels involved in peri-pancreatic aneurysms is technically challenging for the surgeon and as much as 70% of the PDA aneurysms are not detected at surgery^[3].

Non surgical management includes endovascular and percutaneous techniques^[31]. At present endovascular treatment is considered the first choice for embolization of PDA and SMA pseudoaneurysm and the technical success ranges from 56% to 100%^[10,31-33]. Coils, gelfoam, thrombin, glue (N-butyl-cyanoacrylate), stent-graft and Amplatzer vascular plug (AVP, ev3, Plymouth, United States), are different materials for endovascular management and the choice depends on the vascular anatomy of the target lesion and the patient's clinical condition^[34].

Recently, 2 new agents have been used for endovascular treatment of aneurysms and pseudoaneurysms: ethylene vinyl alcohol copolymer (Onyx, Micro Therapeutics, Irvine, CA, United States)^[35] and a multilayer flow modulator stent (Cardatis, Multilayer, Brussels, Belgium)^[32,36].

When antegrade superselective catheterization of the feeding vessel of the PDA pseudoaneurysm fails owing

Table 1 Literature summary of peripancreatic pseudoaneurysms treated with percutaneous thrombin injection

Ref.	Patient age (yr)	Patient sex	Artery	No	Size (mm)	Technique	Needle	Thrombin	Dose (IU)	Immediate result	Adverse effects	Rebleeding	Retreatment	Follow-up (method)
Sparrow <i>et al</i> ^[18]	50	F	GDA	1	30	US-guided	22 G	Bovine	1000	Success	No	No	No	8 wk (US)
Manazer <i>et al</i> ^[19]	42	M	GDA	1	30 × 30	CT-guided	Not specified	Not specified	4000	Success	No	None	No	10 wk (CT)
Armstrong <i>et al</i> ^[21]	Not available	Not available	SMA	1	Not available	US-guided	Not specified	Human	750	Success	Not available	Yes (two times in 6 mo)	Thrombin injection	6 mo (CT)
Geoghegan <i>et al</i> ^[17]	24	M	GDA	1	40 × 35	CT-guided	22 G	Not specified	Not specified	Success	Mild abdominal pain	No	No	1 wk (CT)
Szopinski <i>et al</i> ^[24]	49	F	SMA	1	60 × 44 × 45	US-guided (after failed TCE)	Spinal needle	Bovine	1600	Success	No	No	No	6 mo (US)
Ghassemi <i>et al</i> ^[26]	77	M	SPDA	1	51 × 46	CT-guided (after failed TCE)	22 G	Not specified	1300	Success	No	No	No	4 mo (CT)
Williams <i>et al</i> ^[25]	55	M	IPDA	1	30 × 15	CT-guided	22 G	Bovine	1000	Success	No	No	No	9 mo (CT)
McElean <i>et al</i> ^[9]	46	M	GDA/ PDA	1	25 × 20	US-guided	Not specified	Human	Not specified	Success	No	No	No	3 mo (CT)
De Rosa <i>et al</i> ^[13]	Not specified	Not specified	GDA/ SMA	2	Not specified	CT-guided (after failed TCE)	Not specified	Bovine	4500	Success	Not specified	Yes (24 h)	Not known	15 d (death for PE)
De Rosa <i>et al</i> ^[13]	Not specified	Not specified	GDA/ SMA	1	Not specified	US-guided	Not specified	Human	400	Success	Not specified	No	No	Not specified
Nicholson <i>et al</i> ^[15]	Not specified	Not specified	GDA/ SMA	4	Not specified	CT-guided	Not specified	Not specified	Not specified	Success	Not specified	Yes (multiple)	Thrombin injection	6 mo (with CT)
Laganà <i>et al</i> ^[14]	69	M	GDA	1	50	US-guided + coils	22 G	Bovine	2000	Success	No	Yes (1 mo)	Coils	24 mo
Fankhauser <i>et al</i> ^[16]	Not specified	Not specified	PDA	2	Not specified	US + fluoro-guided	Not specified	Not specified	Not specified	Success	Not specified	No	No	Unknown (mean 524 d)
Present Case Report	82	F	PDA	1	20	US-guided	18 G	Human	1000	Success	No	No	No	3 mo (US)

GDA: Gastroduodenal artery; SMA: Superior mesenteric artery; SPDA: Superior pancreaticoduodenal artery; IPDA: Inferior pancreaticoduodenal artery; PDA: Pancreaticoduodenal artery; US: Ultrasound; CT: Computed tomography; TCE: Transcatheter embolization; G: Gauge; PE: Pulmonary embolism; F: Female; M: Male.

to vessel tortuosity, a retrograde catheterization of the PDA by way of collateral vessels from the GDA could potentially be advantageous.

Unfortunately, in our patient a close stenosis at the origin of the CA did not allowed retrograde catheterization of PDA. As Sutton *et al*^[6] and Kadir *et al*^[7] reported, proximal obstruction of the CA induces consequent compensatory hypertrophy of the PDA and causes hemodynamic alterations that favour the development of PDA aneurysms and pseudoaneurysms^[34,37].

An alternative option to endovascular management of the pseudoaneurysms is the percutaneous embolization by thrombin injection under US or CT guidance.

At first, this method was used for the treatment of iatrogenic pseudoaneurysms of the CFA^[38,39], but there are few reports of treating visceral artery aneurysms and pseudoaneurysms, usually as second procedure after failed transarterial embolization^[9,12,28].

In our case we attempted to identify angiographically the feeding vessel of the pseudoaneurysm, also replacing shape and type of the catheter and changing obliquity of the X-rays beam projection, but superselective catheterization of the feeding branch of the pseudoaneurysm failed.

After exclusion of open surgery approach for high risk-surgery patient, a non-vascular procedure was proposed, by means of percutaneous thrombin injection directly into the pseudoaneurysmal sac.

Thrombin is a clotting factor formed in coagulating form from prothrombin, which hydrolyzes bonds of fibrinogen and allows polymerization of the fibrinogen to form a fibrin clot. The commercially used product is a solution of a sterile protein substance that includes human fibrinogen and artificial aprotinine, which must be mixed with a human thrombin solution in the presence of calcium. After the injection, it acts immediately: the clot is bio-absorbable, and the risk of infection, tissue necrosis or inflammation is minimized.

The first report on the use of thrombin for percutaneous embolization of aneurysms of both peripheral and visceral arteries was in 1986 from Cope *et al.*^[12].

Afterwards this technique was applied to treat several visceral aneurysms and pseudoaneurysms, and type II and type I endoleak following endovascular repair of abdominal and thoracic aortic aneurysms. Potential complications of thrombin use are distal thrombosis, visceral artery occlusion and immunologic reactions^[18]. Given the risk of distal embolization, the use of embolic protection device was suggested^[28]. It is advised to evaluate the free flowing component of the total volume of the pseudoaneurysm, and the quantity of thrombin to prevent the leakage of thrombin from the sac^[20].

The Table 1 presents an overview of the literature where peri-pancreatic pseudoaneurysms were treated by transabdominal thrombin injection US- or CT-guided, along with volume and type of the used thrombin, results, complications and follow up.

In the majority of cases 1000-1500 IU of thrombin were injected into the pseudoaneurysmal sac and the human thrombin is today preferred to bovine thrombin for a minor risk of anaphylactic reaction. In one case additional use of coils was suggested for the embolisation of the afferent artery, to prevent the possible reperfusion of the pseudoaneurysm^[14].

CT usually provides appropriate guide to percutaneous procedure, but US-guide allows real-time evaluation of the amount of thrombin being injected and of the patency of the pseudoaneurysmal sac. Furthermore US-guided injection allows real-time visualization of the needle track, lowering the risk of accidental puncture of bowel, vascular or organs.

Thrombin injection resulted in complete thrombosis and exclusion of the majority of the treated pseudoaneurysmal sacs, even during the follow up imaging control with US or CT scan, although the longest follow-up period was of 24 mo. There were not major complications referred to transabdominal access because of use of small calibre needles, except for our study in which we used an 18 G needle because only an 18 G US-guide was available in emergency. Table 1 shows important risk of early rebleeding of the treated pseudoaneurysm that required at least one additional thrombin injection, re-

ported by some Authors^[13-15,23]. These results indicate the importance of a close post-embolization follow-up with US or CT, required to identify high-risk patients, who need further treatment.

In conclusion, transabdominal thrombin injection could be the first line treatment when the incannulation or identification of the feeding vessel of peri-pancreatic pseudoaneurysm failed, especially in patients not candidate for open surgical approach^[40]. Moreover, in these patients, this treatment could be considered as “bridge-to-surgery” allowing the stabilisation of the ruptured vessel and the further surgical treatment.

COMMENTS

Case characteristics

An 82-year-old woman with cerebral haemorrhage was admitted in emergency with the suspicion of acute bleeding.

Clinical diagnosis

Severe anaemia (haemoglobin drop to 6.0 g/dL) but haemodynamically stable.

Differential diagnosis

Increasing of the cerebral haemorrhage, thoraco-abdominal source of bleeding.

Laboratory diagnosis

Haemoglobin = 6.0 g/dL; haemodynamically stable.

Imaging diagnosis

Contrast-enhanced thoraco-abdominal computed tomography scan showed an oval, rounded mass of 2-cm on diameter within the pancreatic head with slow centrifugal contrast-enhancement, presumed to be a bleeding pseudoaneurysm with a haematoma and a small haemoperitoneum around the spleen.

Pathological diagnosis

Selective angiography of the superior mesenteric artery confirmed a pancreatico-duodenal artery pseudoaneurysm.

Treatment

The patient was treated with 2 mL (1000 IU) of human thrombin percutaneously injected into the pseudoaneurysmal sac under ultrasonography (US) guide.

Related reports

Few reports described peri-pancreatic pseudoaneurysms treated by percutaneous US-guided thrombin injection after failing of the endovascular treatment.

Term explanation

Pancreatico-duodenal artery pseudoaneurysm, or false aneurysm, is a vessel diameter dilatation without all vessel wall layers and is the consequence of a vessel wall disruption limited only by the adventitia or by the surrounding tissues.

Experiences and lessons

Pancreatico-duodenal artery pseudoaneurysm embolization by percutaneous US-guided thrombin injection is feasible after failing of the endovascular treatment, especially in patients not candidate for open surgical approach.

Peer review

This report describes a successful percutaneous US-guided treatment of the rare pancreatico-duodenal artery pseudoaneurysm after a failed endovascular approach.

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