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Endovascular treatment of ruptured pancreaticoduodenal artery aneurysm: The importance of collateral vessels. A case report

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ABSTRACT

INTRODUCTION: True pancreaticoduodenal artery aneurysm occurrence is infrequent, but it is a fatal disease and accounts for accounts for <2% of all visceral aneurysms.
PRESENTATION OF CASE: A 62-year-old man with a two-day history of epigastric pain was admitted at emergency department. CT showed a retroperitoneal haematoma due to a 1.5 cm posterior inferior PDA ruptured aneurysm. Angiography had been conducted immediately: both inflow and outflow of the aneurysm were embolized. Another CT scan had been conducted, which revealed residual flow inside the aneurysm sac fed by small collateral vessels. Sub-selective catheterization was repeated and definitive haemostasis was obtained by embolizing the collateral vessels. Postoperative course was uneventful. CT scan follow-up at 36 months showed no abnormalities.
DISCUSSION: The incidence rate of pancreaticoduodenal artery aneurysm rupture has been estimated to be less than or equal to 65%. In the case of rupture the treatment is challenging and mortality had been reported up to 50%. Endovascular treatment showed superior results as compared to surgical treatment of aneurysms, especially in emergency settings.
CONCLUSION: The authors elucidate the importance of occlusion of inflow and outflow of the aneurysm in conjunction with the occlusion of collateral vessels to avert reperfusion of the sac. Simultaneous handling of celiac axis stenosis is still prone to controversy: no relapse of aneurysm have been reported in patients with celiac axis stenosis at long-term follow-up, simultaneous treatment should be reserved when angiography is alarming for likely hepatic or duodenal ischemia.

1. Introduction

Pancreaticoduodenal artery (PDA) aneurysms are either true or false aneurysms. Pseudoaneurysms can occur if the individual has experienced trauma, surgery, ERCP or pancreatitis. In contrast, true PDA aneurysms have been described in association with occlusion or constriction of the celiac artery that occurs because of median arcuate ligament, arteriosclerosis, or fibromuscular hyperplasia [1,2]. True PDA aneurysm is an infrequent but lethal disease that can result in death in many cases and is responsible for <2% of all visceral aneurysms [1]. PDA aneurysm rupture incidence rate has been determined to be less than or equal to 65% [1–3]. In cases when the rupture occurs, the treatment is considered to be substantially challenging since, in many events, it can lead to death. The literature suggests that the mortality rate for PDA aneurysm rupture is less than or equal to 50% [3,4].

Open surgical repair or endovascular treatment or combining both of these treatments are the three options available to treat ruptured PDA aneurysms. The cases that have been reported to treat PDA aneurysms by using endovascular treatment are limited. Consequently, the goal of this research is to study endovascular treatment as an option for treating this rare disease and discuss the need for concomitant treatment of celiac axis stenosis. The work has been reported according to the SCARE criteria [5].

2. Presentation of case

The present case is related to a 62-year-old man, who does not have any major medical history of trauma, abdominal surgery, or

Abbreviations: PDA, pancreaticoduodenal artery; CT, computed tomography; ERCP, endoscopic retrograde cholangiopancreatography; TAE, transcatheter arterial embolization.

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Fig. 1. Abdominal contrast–enhanced computer tomography (CT) shows a large retroperitoneal haematoma with extravasation from an artery adjacent to the dorsal side of the pancreatic head (white arrow).

Fig. 2. Abdominal contrast–enhanced computer tomography (CT) shows residual extravasation of contrast inside the aneurysm sac after transcatheter embolization of inflow and outflow of the aneurysm with microcoils (white arrow).

pancreatitis and had been admitted to our hospital facility. The patient had a two-day history of epigastric pain and abdominal discomfort. On investigation, the patient's blood pressure measured was 90/50 with the pulse rate 100 beats/min. The hemoglobin concentration was low (8.7 g/dl). On admission, an advanced CT conducted showed that there was a retroperitoneal hematoma. Its size was as big as the pancreatic head and it was observed that 1.5 cm posterior inferior PDA aneurysm was present (Fig. 1). An angiography had been conducted immediately: it documented a posterior inferior PDA aneurysm, mainly fed by the superior mesenteric artery associated with median arcuate ligament syndrome of the celiac axis. The superior mesenteric artery had been subjected to selective catheterization, which was followed by sub-selective catheterization on the aneurysm sac (Progreat 2.7 Fr; Terumo Clinical Supply, Tokyo, Japan). Both inflow and outflow of the aneurysm were embolized with microcoils (Tornado; Cook Medical, Bloomington, USA) to stop the reperfusion of the sac.

Another CT scan had been conducted, which revealed residual flow inside the aneurysm sac fed by small collateral vessels (Fig. 2). Sub-selective catheterization was repeated and definitive hemostasis was obtained by embolizing the collateral vessels with microcoils and a cyanacrylate/lipiodol mixture (Figs. 3 and 4). Postoperative course was uneventful. Consequently, the patient was discharged on the sixth day in good condition. CT scan follow-up at 36 months showed no abnormalities.

3. Discussion

The pancreaticoduodenal cloisters join the superior mesenteric and celiac artery systems by means of the superior pancreaticoduodenal branches off the gastroduodenal artery and the inferior pancreaticoduodenal branches off the superior mesenteric artery [4]. The accurate identification on how the aneurysms are formed is not known but how they progress, has been discussed in literature. It was first discussed by Sutton and Lawton in the 1970s [6]. The true aneurysm forms when the flow in the small caliber vessels increases, which results in local arterial hypertension that causes focal arterial wall weakening and true aneurysm formation.

Although PDA aneurysms represent <2% of all visceral aneurysms, they are known to be fatal and lethal; the incidence rate of PDA aneurysm rupture has been reported to be less than or equal to 65%, compared with a rupture rate of only 3%–9% for splenic artery aneurysms, which have been identified as the most common category of visceral artery aneurysm [1,3]. The rupture is highly fatal but it is not associated with the size of aneurysm. Brocker et al. [4] had investigated more than 90 cases of true PDA aneurysms secondary to celiac stenosis. Their study indicated that the mean aneurysm size found to be 15.2 mm (range 3–40) for the ruptured group and 23.3 mm (range 5–55) for the unruptured group.

Unruptured PDA aneurysms are generally asymptomatic, sometimes presenting with vague abdominal pain related to extrinsic compression of the duodenum or pancreatic and biliary ducts, and diagnosis is incidentally achieved through abdominal CT [3]. The most common clinical presentation of PDA aneurysms is rupture [1,3,4].

In contrast to pseudoaneurysms, ruptured PDA aneurysms rarely present with gastrointestinal hemorrhage, but they usually rupture into the retroperitoneal space and cause acute abdominal pain, which is sometimes associated with symptoms of hematoma such as vomiting or jaundice, whereas most of the patients experience hypotension and shock [1,4]. A mortality rate up to 49.1%, has been reported for ruptured PDA aneurysms [1]. Because of this poor prognosis, the treatment of PDA aneurysms is compulsory for all patients including asymptomatic patients.

Treatment options include surgery (ligation, resection, bypass, pancreaticoduodenectomy) and transcatheter arterial embolization (TAE). In a review of PDA aneurysms by Coll et al., a mortality rate of 20% was reported for patients undergoing surgery, compared with 0% in 13 patients treated by TAE [7]. Similarly, Morita et al. [8] reported a mortality rate of 12.9% for those initially treated with surgery and a mortality rate of 2.1% for those initially treated with embolization.

Indeed, the location of aneurysms of pancreaticoduodenal arcade vessels hinders the surgical extirpation treatment and therefore, it affects the overall medical procedure in a negative manner. In the case of rupture, open surgical repair is particularly difficult, and potentially elevates the morbidity associated with the procedure. Embolization removes the requirement for a challenging surgical procedure, specifically in the case when medical intervention occurs in an emergency setting.

Problems associated with TAE include evaluation of the small feeding artery and achievement of persistent occlusion.

The availability of wires and catheters for super-selective catheterization of small feeding vessels is efficient and effective since it allows accurate localization and treatment of PDA aneurysms. Both inflow and outflow of the aneurysm should ideally be embolized to prevent reperfusion of the sac [9]. However, after

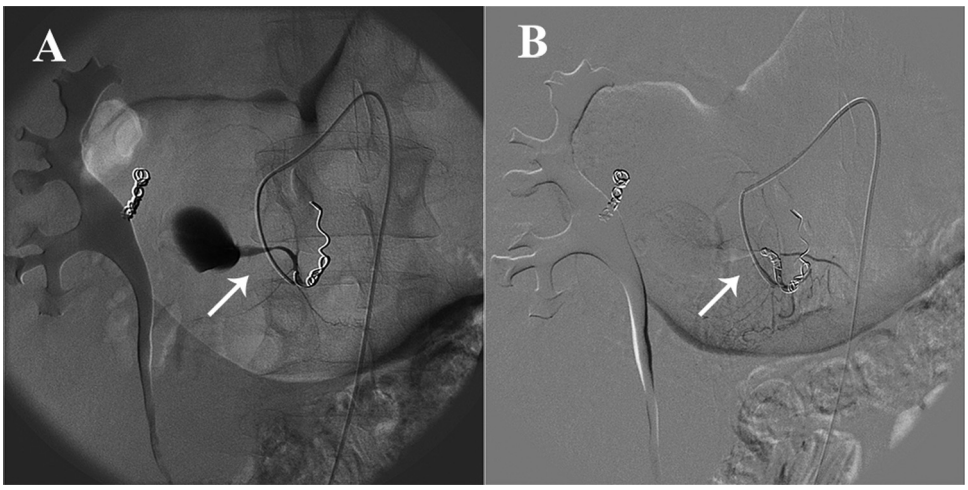

Fig. 3. Super-selective catheter angiography showing collateral vessels that feed the aneurysm sac after embolization of inflow and outflow of the aneurysm with microcoils (A). Definitive haemostasis was obtained by embolizing collateral vessels with microcoils and a cyanacrylate/lipiodol mixture (B).

Fig. 4. Three-dimensional CT image showing luminal stenosis of the celiac axis and definitive occlusion of the aneurysm with microcoils.

embolization, flow to the aneurysm may remain via small collateral vessels, as occurred in the case reported here. Pancreaticoduodenal arcades and other variations of collateralization between the celiac axis and the superior mesenteric artery include the arc of Buhler (an inconstant direct connection between the celiac and the superior mesenteric artery); the dorsal pancreatic artery; and the arcs of Barlow (the collateral pathway within the omentum, between the epiploic arteries of the splenic artery and superior mesenteric artery) [3].

In many cases reported in the literature in which TAE was not successful, the collateral flow to the aneurysm was ignored, leading to persistence of the aneurysm [1]. In the case described herein, TAE was repeated after 24 h to achieve complete occlusion of these small collateral vessels.

In the large series reported by Suzuki et al. [2], the immediate technical success rate of TAE for ruptured PDA aneurysms was 57%. Four of the seven patients had no residual flow to the aneurysm at final angiography, whereas in the other three patients, the flow to the aneurysm experienced a sharp decline and it remained at final angiography, but the remaining arteries were too retracted and convoluted to advance the micro-catheter. Even in the latter cases, all aneurysms were seen to be thrombosed on CT performed 1–2 weeks after embolization, probably due to the small size of the aneurysms (mean size 0.6 cm).

In any case of ruptured PDA aneurysm, TAE should be attempted first if possible. Even if permanent occlusion is not immediately achieved, embolization could be repeated, or in the case of small aneurysms, complete occlusion could be obtained without any other intervention. Finally, in case of residual flow in the aneurysm, temporary control of the bleeding allows a subsequent elective surgical procedure resulting in lower morbidity and mortality rates [10].

The treatment of coexisting celiac axis stenosis remains controversial. Some authors believe that extra treatment may not be required if there is no danger of ischemic dysfunction of the liver and duodenum, whereas others have recommended it to prevent aneurysm recurrence [1,3,4]. Revascularization can be conducted via angioplasty and stenting of the celiac axis in cases of atherosclerotic narrowing or by means of surgical procedures of the median arcuate ligament in patients with median arcuate ligament syndrome. On the other hand, a bypass graft may be created between the celiac/hepatic artery and the superior mesenteric artery.

Recently, Flood et al. [11] studied 14 patients with inferior PDA aneurysms connected with celiac axis stenosis who underwent embolization only or embolization and stenting of the celiac axis. Recurrences of the diseases were not found in the patients after the follow up check up with a time period of 4 years. The researchers also investigated 49 patients reported in the literature. In 40 patients, celiac axis stenosis was not treated together with PDA aneurysm and no recurrence of the aneurysms was revealed

In a series of 93 patients with true PDA aneurysms and celiac axis stenosis/occlusion reviewed by Brocker et al. [4], no recurrence secondary to residual celiac stenosis was reported. Alternatively, 21% of the patients went through treatment of their celiac stenosis, with a mortality rate of 7.6%.

4. Conclusion

Patients with PDA aneurysms should be treated without paying attention to size of the aneurysm and TAE should be a priority in the treatment options. In the case of ruptured aneurysms, TAE should be performed quickly. Considering the morbidity and mortality of the procedure, and that no recurrence of the aneurysm has been described in long-term follow-up, simultaneous treatment of coexisting celiac axis stenosis should be performed only when angiography suggests the potential for hepatic or duodenal ischemia due to stenosis.

Conflicts of interest

No conflicts of interest.

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Ethical approval

The paper is a case report, and therefore does not require ethics approval.

Consent

Written informed consent was obtained from the patient for publication of this Case report and any accompanying images, moreover all details that might disclose the identity of the patient was anonymized.

Author contribution

Gabriele Ricci and Pascale Riu: Concept, design and preparation of manuscript.

Rosaria Mancuso and Carla Di Cosimo: Literature Search and Preparation of Images.

Grazia Maria Attinà and Silvia Trombetta: Acquisition of data, management of the case.

Pasquale Ialongo and Pierluigi Marini: Revision of the Article and Final Approval of the version to be submitted.

Registration of research studies

The case reported is not a first-in-man study and was not registered.

Guarantor

Dr. Gabriele Ricci.

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