Severely Calcified True Aneurysm: A Thought-Provoking Case of Solitary Origin and Postoperative Management

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Conflict of interest:
None declared

Patient:
Male, 70
Final Diagnosis:
Splenic artery aneurysm
Symptoms:
Asymptomatic
Medication:
—
Clinical Procedure:
Surgery and Endoscopy
Specialty:
Surgery

Objective:
Unknown ethiology
Background:
Visceral arterial aneurysms are rare. Most splenic arterial aneurysms (SAAs) are saccular and are in the distal third of the splenic artery. Suggested major causes of SAAs are atherosclerosis, pregnancy, and inflammation. We report the case of a patient who with a SAA extending almost the full length of his splenic artery.

Case Report:
A solitary true aneurysm that extended almost the entire length of the splenic artery was incidentally detected in an asymptomatic 70-year-old male patient with a history of myasthenia gravis and diabetes mellitus. His SAA was severely calcified, but other arteries showed no calcification. The aneurysm had been slightly enlarged toward the celiac artery for 2 years, and aneurysmectomy and splenectomy were performed. Vascular clips were carefully placed at the intact splenic artery without disturbing arterial flows from the celiac artery. Arterial branch from the SAA was ligated at an intact area, and the pancreatic capsule was densely adherent with the calcified aneurysm wall. The pancreas was preserved, although the pancreatic parenchyma was widely exposed during aneurysmectomy. Pathological examination revealed no atherosclerotic changes. Postoperatively, a pancreatic fistula developed, which was treated by placing an intraperitoneal drain and retrograde pancreatic drainage tube. Nevertheless, the intractable pancreatic fistula triggered a bacteriogenic infection, resulting in intraperitoneal abscess. Continuous local lavage via transnasal continuous infusion and endoscopic transgastric drainage was performed, until the fistula closed. He was healthy at 9 months after surgery.

Conclusions:
A SAA that had the rare form and solitary origin was treated. Continuous local lavage has a therapeutic potential for a pancreatic juice-related bacteriogenic complication.

MeSH Keywords:
Aneurysm • General Surgery • Pancreas • Splenic Artery

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Background

Visceral arterial aneurysms are the third most common intra-abdominal aneurysms after aneurysms of the aorta and iliac arteries [1]. Even so, visceral arterial aneurysms are rare, with an incidence of 0.1–0.2% among the global population [2,3]. The splenic artery (SA) is the most frequent site of visceral aneurysms, and SA aneurysms (SAAs) account for approximately 60% of all visceral aneurysms [4]. Beaussier first reported the SAA as an autopsy finding in 1770 [5,6], and thereafter many physicians focused on SAAs. SAAs are divided into 2 forms (saccular or fusiform). Approximately 80% of SAAs are found in the distal third of the SA [7], and have a saccular form [7].

SAAs are classified into 5 types according to their etiology [4]. Although the precise etiology of SAAs remains unclear, atherosclerosis, hypertension, cirrhosis, pregnancy, infection, and inflammation are considered the major causes [4,8–10]. SAAs are more simply categorized into 2 types (i.e., pseudoaneurysm or true aneurysm) [11]. Atherosclerosis generally plays an etiological role in true aneurysms [3,9], whereas infection and/or inflammation are important etiologies for pseudoaneurysms [3].

Isolation of an aneurysmal circulation from any route (i.e., inflow, outflow, collateral pathway, shunt) is a prime principle on which to base SAA treatment [12,13], although a remnant of unincrusted aneurysmal wall is also important. Surgical treatment (e.g., arterial ligation, aneurysmectomy, and splenectomy) and interventional radiology (e.g., covered stent placement and transcatheter arterial embolization) are considered effective [3,14]. Therapeutic indications for asymptomatic and unruptured SAAs are determined by a number if criteria: according to the aneurysm size [1,7,13,15], form and type [13,16], location [11], symptoms [4,7], enlargement [1,7,15], and calcification [15]; if the patient is pregnant [1,4,7] or a young women [7,17]; if portal hypertension is present [1,10]; based on the operative risks [11,17]; and the patient’s clinical status [11,17].

We experienced a solitary SAA that was a severely calcified, nonatherosclerotic, true aneurysm that extended almost the entire length of the SA. We herein describe this thought-provoking case that was treated by surgery and discuss a therapeutic option for the intractable postoperative complication with which we were confronted. We also present a literature review.

Case Report

A 70-year-old male with myasthenia gravis and diabetes mellitus periodically underwent computed tomography (CT), which incidentally detected a severely calcified SAA. He was on oral prednisolone and antidiabetic agents for his comorbidities. The SAA had apparently enlarged over time. Aneurysm size was approximately 15 cm, and the aneurysm was noted to have slowly enlarged toward the celiac artery over 2 years. His primary physician referred him to our department.

Severe calcification of the SA was observed on plain CT (Figure 1A), with no other arteries exhibiting calcification. Dynamic CT revealed a true aneurysm that extended almost the full length of the SA (Figure 1B), rising from the root to the splenic hilum (Figure 1C, 1D). The dorsal pancreatic artery (DPA) branched from the SAA. The common hepatic artery (CHA) and left gastric artery (LGA) were intact (Figure 1C, 1D), and the celiac artery (CA) seemed normal (Figure 2A). The transverse pancreatic, proper hepatic, and gastroduodenal arteries branched from the CHA (Figure 2A, 2B), and DPA branched from the SAA (Figure 2A). The superior mesenteric artery supplied the right hepatic arterial flow. Interventional radiology was excluded from therapeutic options because of risks associated with arterial recanalization, ischemic complications, and cost effectiveness. Hence, we decided on elective surgery.

Initially, arterial branches from the CA i.e., CHA, transverse pancreatic artery (TPA) and LGA, were detected, some with their arterial sheaths (Figure 2C). Thereafter, these arteries were completely skeletonized, removing the arterial sheaths (Figure 2D); i.e., their arterial sheaths were intentionally dissected, and the arterial envelopes were completely exposed to confirm the root of the SA. The proximal side of the patient’s SAA was mobilized, and the root of his SA was considered intact (Figure 3A).

First, 2 clips (Hem-o-lock clip and XL; Teleflex, Wayne, NJ, USA) were placed on the distal side of an intact portion of the SA root. Next, the SAA was cut at the proximal side (Figure 3B). Finally, the third clip was carefully placed at the most proximal side of the intact SA, without disturbing arterial flows to the CHA, TPA, and LGA (Figure 3C, 3D). The abnormal DPA branched from the SAA, and the pancreatic parenchyma was opened to detect an intact portion of the DPA (Figure 4A). The DPA was ligated at an intact portion and then was cut. The pancreatic capsule was densely adherent with the calcified aneurysmal wall, following which the huge SAA was removed from the pancreatic parenchyma (Figure 4B). Normal SA was detected at the splenic hilum and was ligated before venous closure. After inflow control, the splenic vein was sutured using nonabsorbable thread. The pancreas was preserved along with its drainage vein (i.e., the splenic vein), although the pancreatic parenchyma was widely exposed due to the aneurysmectomy. Aneurysmectomy and splenectomy were completed. Drain was intraoperatively placed nearby at the pancreas parenchyma for continuous intraperitoneal drainage after surgery.

Pathology examination revealed that the SAA was composed mainly of calcification. Interestingly, pathological atherosclerosis
was not observed on the pathology evaluation. Cryptogenic or ectopic calcification was suspected, although the severe calcification might have represented a final form of atherosclerosis.

The patient developed a postoperative pancreatic fistula (Figure 4C), and refractory symptoms affected his postoperative course. Amylase level in the drain discharge was routinely checked, and the drain discharge involved pancreatic juice from postoperative day 1. Continuous intraperitoneal drainage was required, and the postoperative pancreatic fistula output was approximately 40 to 80 mL/day. We did not use octreotide to minimize the fistula output. Endoscopic retrograde pancreatic drainage was added on postoperative day 15. Hence, an intraperitoneal drainage via the drain tube and a transpapillary drainage via the endoscopic retrograde pancreatic drainage tube were postoperatively continued. The intractable postoperative pancreatic fistula resulted in abscess formation and localized peritonitis due to a bacteriogenic infection. Continuous local lavage of the abscess cavity was therefore instituted on postoperative day 63 via a transnasal continuous infusion and an endoscopic transgastric drainage (Figure 4D). The abscess cavity, due to the postoperative pancreatic fistula, was continuously washed with a transnasal continuous infusion and simultaneously drained by endoscopic transgastric drainage. A transnasal infusion of saline (40 mL/hour) into the abscess cavity was performed. The refractory postoperative pancreatic fistula finally closed after 1 week of continuous local lavage.
The patient was discharged at postoperative day 73, and was pronounced in good health 9 months after surgery.

**Discussion**

Various etiological factors for SAA have been reported, including angiodysplasia [6], segmental arterial mediolysis [18], portal hypertension [6,8], pregnant or multiparous woman [4,6], atherosclerosis [6,8], essential hypertension [8,19], diabetes mellitus [19], syphilis [6], intracranial aneurysm [19], embolic diseases [6], polyarteritis nodosa [19], α1-antitrypsin deficiency [20], and infective factors and inflammation [6,8].

Interestingly, although the SAA in this case had severe calcification, the pathology evaluation revealed no atherosclerotic changes. Moreover, the aneurysm had a single origin – the SA. Although SAAs had been reported to be related to diabetes mellitus, those patients generally had atherosclerotic changes [19]. We were unable to find any previous reports of SAAs with pathological findings similar to those in our case. Thus, our case of a localized, solitary aneurysm composed only of severe calcification might represent an unusual case of SAA with a rare etiology.

Most SAAs (>90%) remain asymptomatic [21] and are discovered incidentally on imaging studies [22]. Unfortunately,
SAA has a potential for rupture [3,17]. The female/male ratio for SAA is 4:1 [23,24], but the SAAs are more likely to rupture in men [23,24]. The risk of rupture is increased during pregnancy [1,7,17], and is decreased in calcified SAAs [15]. Although the rupture risk is only 3–10% [7,25], the mortality rate is high (20–100%) once rupture occurs [3,14,17,25,26]. As ruptures of both true aneurysms and pseudoaneurysms easily result in life-threatening conditions [27], any aneurysm with rupture or fistula formation requires emergent, aggressive treatment. Isolation of the aneurysmal circulation is a prime principle when treating an SAA. Surgery and interventional radiology are generally employed, as needed. In outpatient's case, interventional radiology was excluded because of risks associated with ischemic complications due to disturbance of arterial flows from the CA, arterial recanalization of the SAA via the DPA, and cost effectiveness. The causal relation between the rupture risk and the size/form of the SAA remains controversial [3,14]. A simple question arose: what was a surgical indication for our case? In our case, the size of SAA had been slowly enlarging over 2 years, and the SAA unfortunately enlarged toward the CA. Therefore, we decided to perform this surgery to preserve arterial flows from the CA, regardless of the size, due to potential risk for rupture.

Historically, a surgical treatment is chosen [3,14], and both robotic and laparoscopic surgical options are currently available [28,29]. From the viewpoint of a minimally invasive treatment, interventional radiology has a large advantage.

**Figure 3.** (A) The root of the SA has an intact portion. The SAA had no effects on the CA. (B) The SAA is cut on the proximal side. Part of the non-circulated wall of the opened SAA remains at the stump. Optimal point for the third clip, nearly at the CA (blue arrow), is carefully estimated beforehand under counter-traction of the SA (red arrow). Branch-patches of the CHA and LGA (yellow line) could be made if arterial reconstruction was required. (C) Finally, the third clip is carefully placed at the most proximal side of the intact SA root under counter-traction of the SA (red arrow). (D) Clip placement does not disturb flow to the CHA, TPA, and LGA from the CA. CA – celiac artery; CHA – common hepatic artery; GDA – gastroduodenal artery; LGA – left gastric artery; PHA – proper hepatic artery; SA – splenic artery; SAA – splenic artery aneurysm; TPA – transverse pancreatic artery.
The mortality rate of elective surgery is >0.5% [30], whereas interventional radiology significantly decreases mortality and morbidity rates [17]. The success rate of the initial interventional radiology for SAA was documented at >90% [31]. Interventional radiology is now applied worldwide [3,14]. However, long-term follow-up after interventional radiology treatment of an aneurysm is required for detecting persistent aneurysmal flow via a collateral pathway and/or shunt [31,32]. Furthermore, postembolization syndrome may develop after interventional radiology treatment for SAA [31] because hepatic, gastric, or bowel ischemia causes serious complications [31]. In our case, interventional radiology was excluded from therapeutic options based on 3 factors: 1) the technical difficulty of achieving complete isolation of the SAA because of the coexistence of its arterial branches (e.g., the DPA); 2) the risk of postembolization syndrome due to unexpected disturbance of arterial flow into other organs (especially flow from the CA); and 3) the lack of cost effectiveness. Although arterial flow from the CHA and LGA were not affected in our case, even after clipping procedures, we were ready to undertake arterial reconstruction of the SAA.

**Figure 4.** (A) An abnormal DPA branches from the SAA, and the pancreatic parenchyma is explored to detect an intact portion of the DPA. The DPA is ligated at an intact portion and then cut. (B) Severely calcified aneurysmal wall and dense pancreatic capsule coalesce, and the SAA is removed from the pancreatic parenchyma. (C) The patient suffered from refractory symptoms due to a postoperative pancreatic fistula, and enhanced CT findings revealed an intraperitoneal abscess related to the PPF (orange arrows). (D) Pancreatic drainage tube was placed in a retrograde fashion (green). The abscess cavity due to the PPF (orange area) was continuously washed with a transnasal continuous infusion (red) and simultaneously drained by endoscopic transgastric drainage (blue). A collection rate of >80% for a saline wash of the abscess cavity was confirmed during endoscopic cannulation. Thereafter, a transgastric drainage tube was placed. A transnasal infusion tube was also placed using the same transgastric orifice (dotted line), and a continuous flow of saline (40 mL/hour) into the abscess cavity was established. The collection rate of a saline injection into the abscess cavity was decreased to approximately 60%, after a transgastric drainage. Amylase level in the lavage fluid was decreased to within 100 IU/L. The intractable, refractory PPF disappeared after continuous local lavage induction. CT – computed tomography; DPA – dorsal pancreatic artery; PPF – postoperative pancreatic fistula; SA – splenic artery; SAA – splenic artery aneurysm.
the CHA and LGA using a branch-patch technique (i.e., branch-patch of the CHA and LGA anastomosed to the aorta) [33] if aneurysmal changes reached the CA (Figure 3B). Retrospectively, however, we thought that intractable postoperative pancreatic fistula could easily cause fatal complications (e.g., anastomotic rupture), if an arterial anastomosis was created.

Postoperative pancreatic fistula is an intractable pancreatic juice-related complication, and a subsequent bacteriogenic infection seriously worsens the patient condition. Although the severely calcified aneurysmal wall and pancreatic capsule coalesced in our patient, the pancreatic parenchyma was injured during aneurysmectomy. Moreover, the pancreatic parenchyma was cut into and explored to ensure that the DPA had been ligated correctly at the intact arterial wall. We speculated that these procedures might explain the refractory postoperative pancreatic fistula. In the therapeutic strategy for acute severe pancreatitis, continuous local lavage is currently accepted as an effective treatment option [34–37]. In our case, continuous local lavage was performed via a transnasal infusion and a transgastric drainage, with the pancreatic drainage tube placed in a retrograde fashion (Figure 4D). After continuous local lavage induction, the intractable postoperative pancreatic fistula with bacteriogenic infection disappeared rapidly. Historically, only surgical approaches have been recommended for treating necrotizing and infected pancreatitis [38,39]. Today, however, continuous local lavage is currently accepted as an effective treatment for severely infected pancreatitis [34–37]. We suggest that continuous local lavage may have a therapeutic potential even for pancreatic juice-related complications, including postoperative pancreatic fistula. We report our patient case as a thought-provoking SAA case, and hope our report will be informative for further developments in SAA treatment.

Conclusions

A solitary and localized SAA involved almost the entire length of the splenic artery, and was enlarging towards to the CA. In such cases, aneurysmectomy with splenectomy is preferable over interventional radiology. As the SAA was adherent to the pancreatic capsule, the risk of development of pancreatic fistula postoperatively was high. Postoperative continuous local lavage offers good therapeutic potential for a pancreatic juice-related bacteriogenic complication, if it develops.

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Ethical approval

This report was approved by the Institutional Review Board of Shiga General Hospital, Moriyama, Japan.

Conflicts of interest

None.

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