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CASE REPORT

Pancreatic arteriovenous malformation treated with transcatheter arterial embolization: Two case reports and review of literature

Sang Hoon Shin, Chol Kyoon Cho, Sung Yeol Yu

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Abstract

BACKGROUND

Various treatment methods are available for the treatment of pancreatic arteriovenous malformation (P-AVM); however, there are no established treatment options for asymptomatic P-AVM.

CASE SUMMARY

A 47-year-old and a 50-year-old male patients sought treatment for P-AVM in the pancreas, which was incidentally detected during routine abdominal computed tomography and magnetic resonance imaging conducted as part of a health check-up. They underwent transcatheter arterial embolization (TAE), and over the course of a 9-year follow-up period, the AVM did not worsen and was asymptomatic.

CONCLUSION

TAE can be considered as an alternative treatment option for P-AVM in selective cases where patients are asymptomatic or have a high surgical risk.

Key Words: Pancreatic arteriovenous malformation; Transcatheter arterial embolization; Surgical treatment; Asymptomatic; Angiography; Case report

Core Tip: Pancreatic arteriovenous malformation (P-AVM) is a rare condition characterized by symptoms like gastrointestinal bleeding and abdominal pain, with some cases being asymptomatic. Surgical intervention is commonly considered for symptomatic cases, but the treatment of asymptomatic P-AVMs is not well-established. Previous studies have reported various methods, including surgery and transcatheter arterial embolization (TAE). In selective cases, particularly for asymptomatic or high surgical risk patients, TAE may be an effective and safe treatment option.

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INTRODUCTION

Pancreatic arteriovenous malformation (P-AVM) is a rare condition characterized by abnormal blood flow resulting from anastomoses between arteries and veins[1]. Since its initial description by Halpern *et al*[1] in 1968, fewer than 100 cases of P-AVM have been reported in the literature[2]. P-AVM can present with various symptoms including abdominal pain, gastrointestinal (GI) hemorrhage, and jaundice, or it may be discovered incidentally without any specific symptoms[3].

P-AVM diagnosis can be confirmed with various imaging modalities such as contrast-enhanced computed tomography (CT), magnetic resonance imaging (MRI), and angiography. In contrast-enhanced CT and MRI scans, imaging findings often show the presence of dilated and tortuous feeding arteries that supply the abnormal vascular network within the pancreas[4]. Additionally, angiography is particularly useful for identifying the abnormal vascular network associated with P-AVM, with the early filling of draining veins (*e.g.*, the portal vein) indicating the presence of P-AVM[5].

Surgery is often considered as the primary treatment option for symptomatic patients with P-AVM; nevertheless, other treatment modalities such as transcatheter arterial embolization (TAE), radiation therapy, and even conservative management have been considered by researchers[2,6-8].

Here, we present a report of two cases of asymptomatic P-AVM treated with TAE and an additional literature review on P-AVM.

CASE PRESENTATION

Chief complaints

Case 1: A 47-year-old male patient came to our hospital for treatment due to suspicion of P-AVM, which was identified during a health checkup at a different medical institution.

Case 2: A 50-year-old male patient came to our hospital for treatment due to suspicion of P-AVM, which was identified during a health checkup at a different medical institution.

History of present illness

Case 1: The patient, who was asymptomatic, underwent a routine health check-up at a local hospital. The laboratory tests performed during the check-up did not show any abnormalities. However, as the abdominal ultrasound showed abnormal findings in the pancreas, an abdominal CT was performed. Additionally, to obtain more detailed information, MRI was conducted, which detected P-AVM. For the treatment this lesion, the patient visited our hospital.

Case 2: The patient, who was asymptomatic, underwent a routine health check-up at a local hospital. The abdominal ultrasound showed abnormal findings in the pancreas, which led to the decision to perform MRI. Subsequently, abdominal CT was conducted, which revealed the presence of pancreatic neck and body AVM. The patient visited our hospital for the treatment of this lesion.

History of past illness

Case 1: The patient had hypertension (HTN) and chronic kidney disease (CKD) detected 4 months ago. HTN was well-controlled (mean systolic blood pressure 140 mmHg) with medication. CKD was diagnosed as advanced renal failure (Cr 5.1 mg/dL, eGFR 11.79 mL/min/1.73 m^2), stage 5.

Case 2: The patient was diagnosed with diabetes mellitus 1 year ago, which was effectively managed with medication, resulting in well-controlled blood sugar levels.

Personal and family history

Cases 1 and 2: The patients had no family history.

Physical examination

Cases 1 and 2: The patients' abdomen was soft upon palpation, without any abnormalities.

Laboratory examinations

Cases 1 and 2: The laboratory tests were normal.

Imaging examinations

Case 1: Abdominal contrast-enhanced CT and MRI revealed a well-enhanced hypervascular lesion of around 4.0 cm located in the pancreatic head. The portal vein was filled early with arterial blood during the arterial phase, leading to the suspicion of P-AVM (Figure 1).

Case 2: Abdominal contrast CT and MRI scans showed a 5.2 cm × 4.0 cm dilated and twisted hypervascular lesion in the pancreas neck and body, with strong enhancement of the vessels and arterial blood presence in the portal vein during the arterial phase (Figure 2).

Further diagnostic work-up

Case 1: Before intervention, the patient underwent follow-up laboratory tests. Based on the results, the patient had slightly elevated amylase (136 U/L) and lipase (109 U/L) levels, along with an increased CRP level (5.73 mg/dL); however, serum tumor marker levels were normal.

Case 2: Before intervention, all laboratory tests including tumor markers were within the normal range. Additionally, an irregular cystic lesion measuring approximately 2.0 cm was identified in the neck and body of the pancreas during abdominal endoscopic ultrasonography (Figure 3).

FINAL DIAGNOSIS

Cases 1 and 2

P-AVM.

TREATMENT

Case 1

Considering the patient's asymptomatic status and high surgical risk, we decided to perform TAE. Angiography of the SMA, celiac axis, and gastroduodenal artery was conducted, showing hypervascular staining of the tumor of the pancreatic head. Superselective catheterization of the gastroduodenal artery showed multiple feeding branches originating from the pancreaticoduodenal arcade. The pancreaticoduodenal arcade and dorsal pancreatic artery were treated using interlock coils. Interlock coils are specialized devices designed specifically for the purpose of occluding blood vessels (Figure 4).

Case 2

Considering the patient's asymptomatic status and diffuse lesion of the pancreas, we decided to perform TAE. Angiography of the SMA, celiac axis, and common hepatic artery was performed, which showed hypervascular staining in the pancreatic neck and body. The feeding branches from the proper hepatic artery and dorsal pancreatic artery were then embolized using interlock coils (Figure 5).

OUTCOME AND FOLLOW-UP

Case 1

A follow-up abdominal computed tomography angiography performed on the 5th day after TAE showed no significant change in the size of the hypervascular lesion in the pancreatic head (Figure 6). The patient had persistent symptoms such as fever, chills, and upper abdominal pain beginning on the 5th day after the procedure. An esophagogastroduodenoscopy was performed to further evaluate the condition of the patient, which showed evidence of ischemic duodenitis and atrophic gastritis (Figure 7). After TAE, the patient received conservative treatment for approximately 1 week, during which the symptoms were gradually improved, and the patient was discharged. Conservative treatment included nutrition support, proton pump inhibitor medication, pain control, and hydration.

After discharge, the patient underwent regular follow-ups, including abdominal CT and laboratory tests, which were initially performed every 3 to 6 mo for 2 years before subsequently transitioning to an annual follow-up schedule.

During the 9-year follow-up period, the AVM showed a significant improvement in hypervascularity, with the lesion size decreasing from 3.7 cm to 2.3 cm. The patient remained asymptomatic (Figure 8).

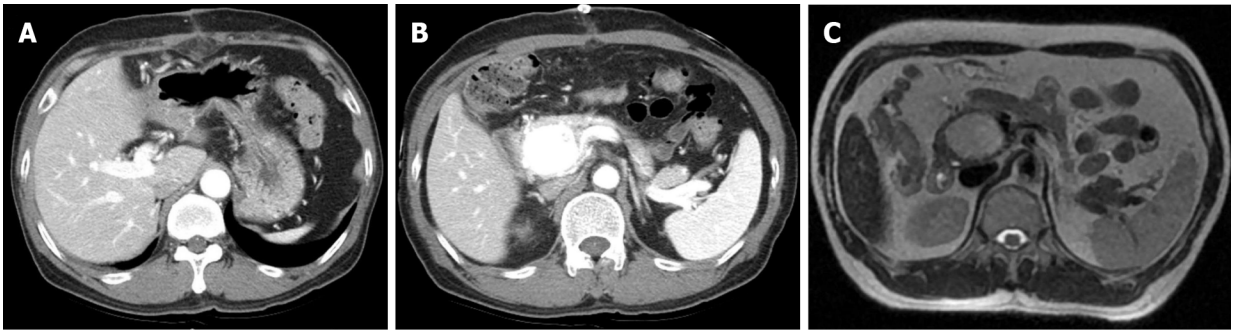

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Figure 1 Abdominal computed tomography and magnetic resonance imaging findings before transcatheter arterial embolization (case 1). A and B: Abdominal computed tomography images demonstrating early arterial filling of the portal vein (A) and a 3.7 cm hypervascular lesion in the pancreas head (B); C: Additionally, magnetic resonance imaging image showing a similar lesion.

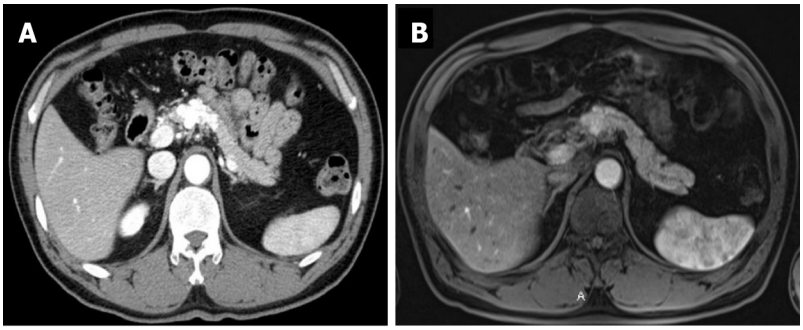

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Figure 2 Abdominal computed tomography and magnetic resonance imaging before transcatheter arterial embolization (case 2). A: Computed tomography image showing a 5.2 cm × 4.0 cm enhancing tortuous, tubular hypervascular lesion in the pancreas neck, and body; B: Magnetic resonance imaging image showing a 2 cm multilobulated and irregular hypervascular lesion in the pancreatic neck and body with peripancreatic infiltration.

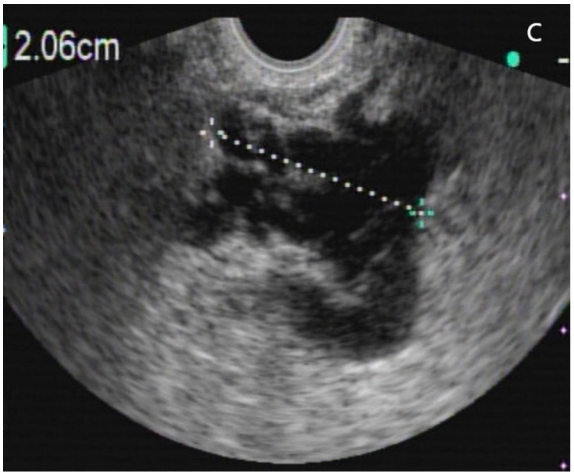

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Figure 3 Abdominal endoscopic ultrasonography (case 2). Abdominal endoscopic ultrasonography showing the presence of an approximately 2.0 cm cystic lesion.

Case 2

A follow-up CT scan on the 4th day after TAE showed reduced contrast enhancement and hypervascularity (Figure 9). However, during hospital stay, the symptoms of the patient gradually improved, and the patient was discharged.

After discharge, the patient underwent regular follow-ups, including abdominal CT and laboratory tests, which were initially performed every 3 to 6 mo for 2 years before subsequently transitioning to an annual schedule.

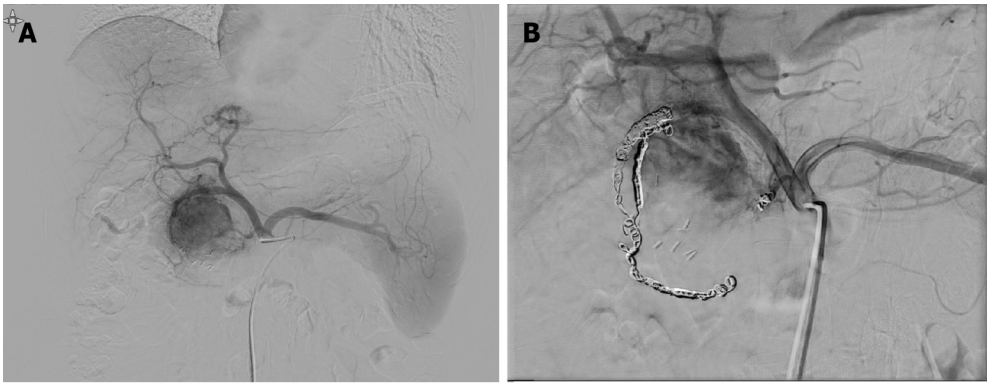

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Figure 4 Angiography and transcatheter arterial embolization (case 1). A: Angiographic images revealing multiple feeding branches supplying blood flow to the nidus; B: While the interventional image shows the use of interlock coils to occlude the pancreaticoduodenal arcade and dorsal pancreatic artery.

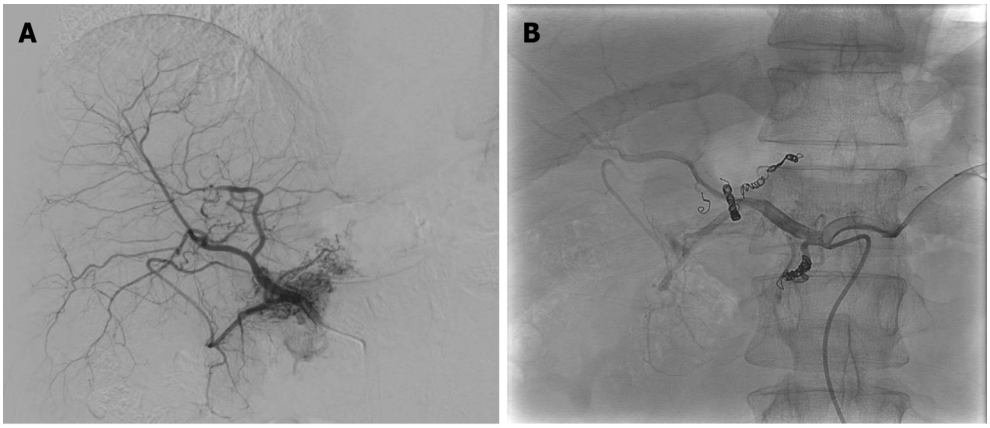

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Figure 5 Arteriography and transcatheter arterial embolization (case 2). A: Angiographic images of feeding branches from the proper hepatic artery and the dorsal pancreatic artery; B: The feeding branches are then treated with interlock coils to block the blood supply to the abnormal vessels in the pancreas.

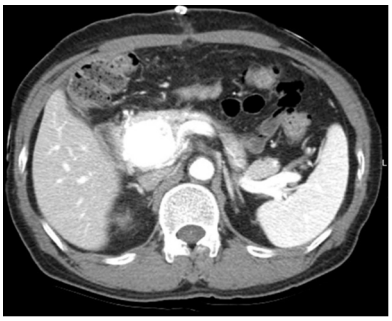

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Figure 6 Abdominal computed tomography performed on day 5 after transcatheter arterial embolization (case 1). Post-transcatheter arterial embolization image showing improvement in arteriovenous malformation and a slight reduction in peripancreatic hypervascular lesions.

During the 3rd year of follow-up, no significant increase in the hypervascularity of the P-AVM was observed; however, the size of the lesion changed from 5.2 cm × 4.0 cm to 5.7 cm × 3.3 cm. No significant changes were observed in subsequent follow-up examinations over the next 9 years (Figure 10). During the 9-year follow-up period, the patient remained asymptomatic, and there were no significant changes in the size of the lesion in the pancreatic head, which remained at 6.0 cm × 3.0 cm.

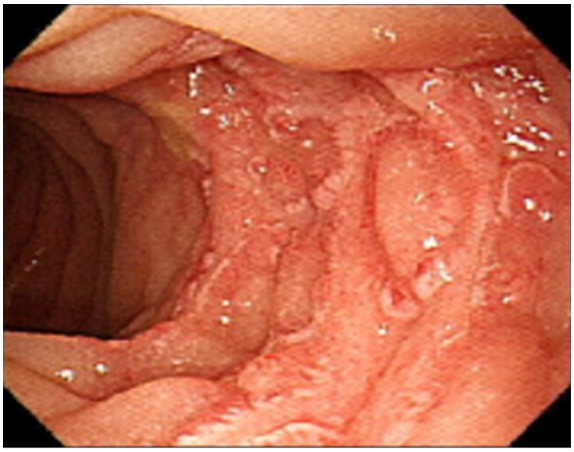

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Figure 7 Esophagogastroduodenoscopy performed on day 10 after transcatheter arterial embolization (case 1). The presence of ischemic duodenitis is evident, characterized by segmental circumferential erythematous uneven mucosal changes with nodularity.

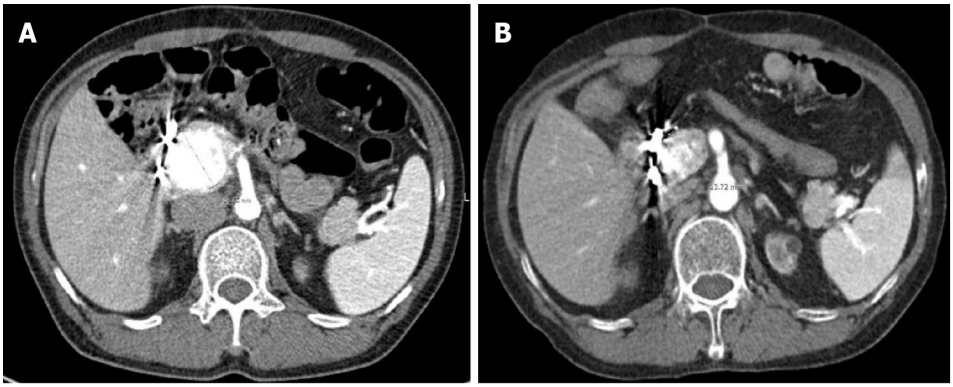

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Figure 8 Follow-up abdominal computed tomography scans three months and nine years after the procedure (case 1). A: The computed tomography (CT) scan after three months revealed a 3.7 cm hypervascular lesion; B: While the CT scan after nine years showed a reduction in the size of the hypervascular lesions to 2.3 cm.

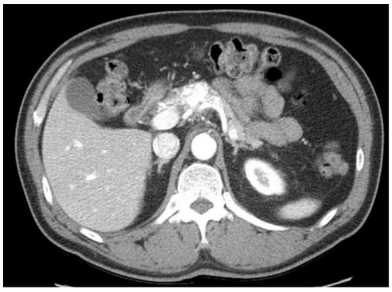

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Figure 9 Abdominal computed tomography findings before transcatheter arterial embolization (case 2). Post-transcatheter arterial embolization computed tomography scan performed on the 4th day revealed reduced contrast enhancement.

DISCUSSION

P-AVM is a rare condition characterized by the abnormal connection between the arterial and portal systems in the pancreas, resulting in tumor formation and blood flow abnormalities[8]. Histological examinations usually identify dilated and twisted blood vessels within the pancreatic parenchyma as well as collections of irregularly tortuous blood vessels with thick walls[3,9,10].

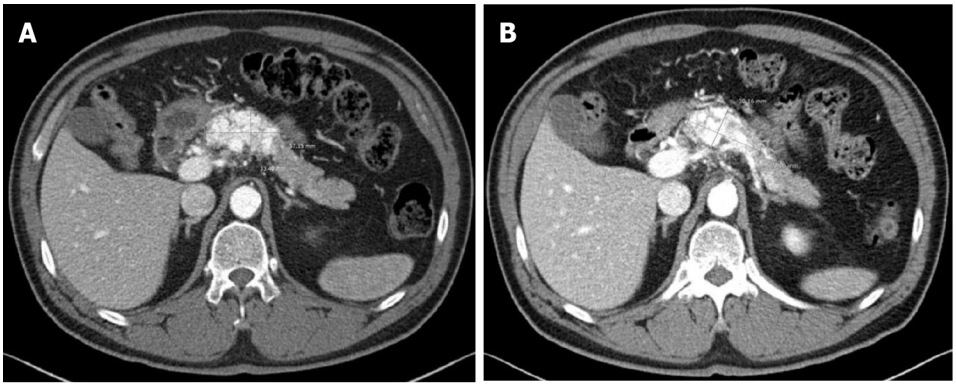

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Figure 10 Follow-up abdominal computed tomography scan. A: On the three-year follow-up computed tomography scan, there was no significant change in the lesion size, which measured 5.7 cm × 3.3 cm; B: On the nine-year follow-up scale, the lesion size remained at 6.0 cm × 3.0 cm.

Chou *et al*[11] (2013) conducted a study on 89 patients with P-AVM, which showed that it was more common among males (85.4%) than among females (14.6%). The median age of the patients in their study was 50 years. Furthermore, Wu *et al*[3] (2021) reported a male predominance (24 males, 2 females) among P-AVM patients, with an average age at diagnosis of 51.5 years.

AVM can occur in any part of the GI tract; however, the incidence of P-AVM is very low, accounting for only 0.9% of all AVM cases[11-15]. Several studies have reported that the majority of P-AVMs (48.3%-62.3%) are located in the pancreatic head, followed by the pancreatic body and tail and the entire pancreas[10,12,16].

Common symptoms of P-AVM include abdominal pain and GI bleeding; however, acute pancreatitis, portal HTN leading to ascites, esophageal variceal rupture, and severe GI bleeding are also associated with P-AVM[10]. Chou *et al*[11] (2013) showed that GI bleeding was the most common symptom (47.2%), followed by epigastric pain (46.1%) and back pain (9%), with a small percentage of patients (4.5%) being asymptomatic.

CT, MRI, and angiography are commonly used to diagnose P-AVM as they can detect dilated and tortuous feeding arteries supplying the abnormal vascular network in the pancreas[4]. Angiography can also provide a detailed visualization of blood vessels, with the early filling of veins confirming abnormal blood flow and the presence of P-AVM[5].

Angiography is valuable for obtaining detailed information about the extent and characteristics of P-AVM, including multiple lesions, localization, and spread. It also plays an important role in treatment planning by identifying feeding arteries and determining the optimal target for embolization[17,18]. According to Wu *et al*[3] (2021), Color Doppler ultrasonography is a useful diagnostic tool for P-AVM, particularly in identifying the characteristic "mosaic sign" of vascular malformation associated with P-AVM.

Yakes *et al*[19] (2014) developed a categorization system for AVM based on the inflow arteries, outflow veins, and AVM type. This classification includes five types of AVM. Type 1 has a single arterial input without early venous drainage. Type 2 involves multiple arterial inputs with no early venous drainage. Type 3 exhibits early venous drainage with a single draining vein (subdivided into Type 3a with low flow and a small-diameter draining vein and Type 3b with high flow and a large-diameter draining vein). Type 4 has multiple arterial inputs with early venous drainage. Our AVM cases showed multiple feeding branches and vein outflow on contrast-enhanced abdomen CT and arteriography, which could be classified as Type 2 AVMs.

P-AVM is normally treated with surgical resection or TAE; however, other treatment options such as radiotherapy, trans-jugular intrahepatic portosystemic shunt (TIPS), and conservative management may also be considered[2].

Surgical resection is considered as a curative treatment modality for P-AVM. However, it is important to carefully evaluate the potential risks associated with the procedure. Pancreatectomy for AVM carries the risk of significant bleeding during or after the operation, and the invasiveness of the procedure may lead to the development of diabetes as a potential side effect. Therefore, a thorough assessment of these risks is crucial when considering pancreatectomy as a treatment option for P-AVM[2].

Chou *et al*[11] (2013) reported that primary treatments for P- AVM were surgery (43.8%) and TAE (11.2%). A smaller proportion of cases involved a combination of surgery and TAE (10.1%), and a minority of patients were treated with radiotherapy (2.2%). However, a significant percentage of P-AVM patients (29.2%) were managed through conservative treatment without any intervention[8].

TAE is less invasive for P-AVM management but carries a higher risk of recurrence and potential complications requiring additional treatments. Hakoda *et al*[20] (2022) reported that TAE may be associated with a higher risk of AVM recurrence and the potential for embolization of other portal branches because of the migration of embolic agents. Additionally, achieving complete embolization may be difficult as the AVM is often supplied by multiple feeding arteries. Wu *et al*[3] (2021) found that 57.7% of the 26 patients in their study who underwent TAE for P-AVM were successfully treated, whereas the remaining 42.3% of patients required additional surgical intervention. They also reported that surgical resection may be the most effective treatment option for P-AVM, and alternative approaches such as TAE, TIPS, and radiotherapy may not be able to effectively manage the associated complications.

Table 1 Reported cases of pancreatic arteriovenous malformation treated with transcatheter arterial embolization

Ref.	Year	No. of cases	Age	Sex	Initial main symptoms	Location	Symptoms or complications after TAE	Additional treatment
Gomes *et al*[22]	1982	1	52	M	Abdominal pain	Head	GI bleeding	PD
Kato *et al*[23]	1991	1	60	M	Asymptomatic	Body or/and tail	None	None
Ishikawa *et al*[24]	1993	1	66	M	Abdominal pain	Head	None	None
Hirai *et al*[25]	1995	1	67	M	Asymptomatic	Body or/and tail	Unknown	Radiation
Hayashi *et al*[8]	1998	1	45	M	GI bleeding	Head	GI bleeding	TIPS
Uda *et al*[26]	1999	1	48	M	Abdominal pain	Head	Unknown	PPPD
Iwashita *et al*[27]	2002	1	58	M	GI bleeding	Head	None	None
Sato *et al*[7]	2003	1	60	M	GI bleeding	Head	GI bleeding	Radiation
Hosogi *et al*[28]	2006	1	45	M	GI bleeding	Head	Unknown	PPPD
Ogawa *et al*[29]	2009	1	54	M	Asymptomatic	Body or/and tail	None	None
Gincul *et al*[30]	2010	1	55	M	Abdominal pain	Head	None	None
Charalabopoulos *et al*[12]	2011	1	64	F	Abdominal pain	Body or/and tail	Abdominal pain	None
Sharma *et al*[5]	2011	1	26	M	Abdominal pain	Head	GI bleeding	PD
Qayed *et al*[31]	2011	1	47	M	GI bleeding	Head	GI bleeding	PPPD
Grasso *et al*[32]	2012	1	48	M	GI bleeding	Head	Duodenal ulcers	None
Song *et al*[6]	2012	2	44 (1), 46 (1)	M (2)	Abdominal pain (2)	Head (2)	Unknown (2)	PPPD (2)
Arora *et al*[33]	2013	1	37	Male	Abdominal pain	Head	GI Bleeding	PD
Uojima *et al*[34]	2014	1	49	Male	Abdominal pain	Head	None	None
Tatsuta *et al*[35]	2014	1	57	Male	Abdominal pain	Head	None	None
Cassinotto *et al*[36]	2015	1	56	Male	Abdominal pain	Head	Duodenal ulcers	None
Fukami *et al*[37]	2015	1	50	Male	Abdominal pain	Head	None	PPPD
Vidmar *et al*[38]	2016	1	54	Male	Abdominal pain	Head and body	None	None
Kohan *et al*[39]	2017	1	46	Male	Abdominal pain	Body or/and tail	None	None
Gupta *et al*[14]	2018	1	60	Male	Abdominal pain	Head	None	PPPD
Yoon *et al*[2]	2020	1	43	Male	Abdominal pain	Body or/and tail	None	None
Marcelin *et al*[21]	2022	5	61.1 (43-79)	M (1), F (4)	GI bleeding (3), abdominal pain (1), ascite (1)	Head (5)	GI bleeding (1), none (4)	None (4), PPPD (1)
Current studies	2023	2	56 (1), 59 (1)	M (2)	Asymptomatic (2)	Head (1), body (1)	Ischemic duodenitis (1), none (1)	None (2)

GI: Gastrointestinal; M: Male; F: Female; TAE: Transcatheter arterial embolization; PD: Pancreaticoduodenectomy; PPPD: Pylorus-preserving pancreaticoduodenectomy.

Other studies have investigated the effectiveness of TAE for P-AVM. Marcelin *et al*[21] (2022) conducted a study with 7 patients to evaluate the efficacy and safety of TAE for patients with P-AVM. A total of 5 of the 7 patients underwent TAE for symptomatic P-AVM, with 80% of patients experiencing complete resolution of P-AVM symptoms. Among patients with incomplete embolization, 25% of patients required additional surgery, and 75% of patients responded well to conservative treatment. They observed the progressive regression of P-AVM with no symptom recurrence despite incomplete embolization. Therefore, they suggested that TAE may be considered as a safe and effective treatment option for selected cases of symptomatic P-AVM, with surgical intervention reserved as a secondary option[20].

We have conducted a case review focusing on TAE as a treatment option for P-AVM, which included our cases. Of the 33 cases included in our review, the majority of cases involved males (29 cases, 87.9%). The most common initial symptom was abdominal pain, which was reported in 18 cases (54.5%). GI bleeding was observed in 9 cases (27.3%). There was 1 case (3%) of ascites, and the remaining 5 cases (15.2%) were asymptomatic. In 26 cases (78.8%), the pancreatic head was the predominant location for P-AVM, and the remaining 7 cases (21.2%) involved either the body or tail region. After TAE, most patients (17 cases, 51.5%) showed no post-procedure symptoms or complications; however, there were GI bleeding in 7 cases (21.2%), duodenal ulcers and duodenitis in 3 cases (9.1%), and abdominal pain in 1 case (3%). The remaining 5 cases (15.2%) had unknown complications (Table 1). These findings offer valuable insights into the clinical characteristics and outcomes of TAE as a treatment option for P-AVM[3].

This study has some limitations, such as a small number of included patients and the use of a retrospective design. The limited sample size might not fully represent the entire population of patients with P-AVM. To strengthen the evidence on the effectiveness of TAE for P-AVM, prospective studies will be necessary in the future. Well-designed prospective studies can provide more reliable evidence on the outcomes of TAE. These studies may potentially involve a larger patient cohort with a longer follow-up period, allowing for a more comprehensive evaluation of treatment efficacy.

Despite these limitations, this study provides valuable insights into the management of P-AVM with TAE. The findings could contribute to a more comprehensive understanding of the efficacy and safety of TAE as a treatment option for P-AVM, ultimately leading to improved patient outcomes.

CONCLUSION

There are currently no established treatment guidelines for P-AVM; however, there are various treatment options available, including surgical resection and TAE. Surgical resection is the mainstay of treatment for symptomatic patients. Nevertheless, TAE can be considered as an alternative treatment option in selective cases where patients are asymptomatic or have a high surgical risk.

FOOTNOTES

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