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CASE REPORT

Gastrointestinal Bleeding Due to the Rupture of Splenic Artery Caused by Pancreatic Carcinoma: A Case Requiring Repeated Transcatheter Arterial Embolization in a Short Period of Time

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Abstract:
In this report, we present a case of gastrointestinal bleeding due to splenic artery rupture, which required repeated transcatheter arterial embolization (TAE) within a short period of time. A 75-year-old man with pancreatic carcinoma was transported to our hospital with active hematemesis and vital signs consistent with shock. Contrast-enhanced computed tomography images showed a pancreatic tumor that had caused a pseudoaneurysm of the splenic artery to rupture. The pseudoaneurysm was embolized using only an N-butyl-2-cyanoacrylate (NBCA) and lipiodol mixture. However, hematemesis with signs of shock recurred 13 h later, and angiography showed rebleeding from the origin of the splenic artery. The splenic artery was subsequently embolized using an NBCA and lipiodol mixture. Repeated TAE finally controlled the hemorrhage; however, asymptomatic splenic infarction and hepatic infarction occurred due to nontarget embolization.

Keywords:
splenic artery pseudoaneurysm, rebleeding, N-butyl-2-cyanoacrylate, transcatheter arterial embolization

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Introduction

Pancreatitis often accompanies pancreatic cancer and may affect adjacent arteries, such as the splenic artery, leading to arterial irregularities and pseudoaneurysm formation due to arterial wall disruption in the presence of free lipolytic and proteolytic enzymes in the pancreatic juice [1]. Bleeding from a peripancreatic pseudoaneurysm can be fatal and requires urgent transcatheter arterial embolization (TAE) [2]. Although TAE has become the standard treatment option for a pseudoaneurysm, if not performed appropriately, it can result in rebleeding shortly afterward [3].

TAE using only an N-butyl-2-cyanoacrylate (NBCA) and lipiodol mixture is a quick, feasible, and effective treatment for pseudoaneurysms with multiple collateral sources or small tortuous target vessels [4]. However, to avoid nontarget embolization, TAE using only an NBCA and lipiodol mixture to the main trunk of major visceral pseudoaneurysms, such as the splenic artery, is not usually performed.

In this report, we present a case of gastrointestinal bleeding in a critically ill patient with pancreatic carcinoma complicated by pseudoaneurysm formation, which required repeated TAE using only an NBCA and lipiodol mixture; however, due to nontarget embolization, asymptomatic splenic infarction and hepatic infarction occurred.

Case Report

Prior to the publication of this case report, the patient provided written informed consent.

A 75-year-old man with pancreatic carcinoma invading the duodenum and surrounding vessels from the celiac artery to the splenic artery was transported to our hospital with active hematemesis and vital signs consistent with shock vital. On arrival, his vital signs and laboratory data were as follows: blood pressure, 83/67 mmHg; heart rate, 95 beats/min; hemoglobin level, 8.3 g/dL; platelet count, 14.5 $\times$ 10^4/μL; fibrinogen level, 282 mg/dL; prothrombin time,

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12.9 s; and activated partial thromboplastin time, 25.6 s. Despite fluid resuscitation, his condition deteriorated, and his vital signs became unstable; his blood pressure had dropped to 34/23 mmHg at its lowest. Contrast-enhanced computed tomography (CECT) images showed a large necrotic pancreatic tumor involving the proximal portion of the splenic artery with a large pseudoaneurysm with extravasation of contrast material into the necrotic tumor and then into the duodenum via the fistula (**Fig. 1** and Supplementary Video 1). The tumor also encased the common hepatic artery and the celiac axis. Nevertheless, the splenic artery did not have direct contact with the lumen of the gastrointestinal tract. Based on the clinical status and CECT findings, an emergent TAE was performed immediately after the arrival of the on-call interventional radiologist (**Fig. 2**).

A 5 F sheath (Radifocus Introducer IIH, Terumo, Tokyo, Japan) was inserted into the right femoral artery. Celiac angiography was performed using a 5 F shepherd hook catheter (Hanako Medical Co. Ltd., Saitama, Japan), which revealed a pseudoaneurysm of the splenic artery (**Fig. 2a**). Celiac angiography also showed irregular narrowing and dilatation of the proximal portion of the splenic artery and the common hepatic artery, which represented tumor invasion. The pseudoaneurysm was catheterized with a 2.0 F tip microcatheter (Bobsled Allorounder; Piolax Inc., Kanagawa, Japan) and a 0.014-inch steerable guidewire (Labyrinth Noah; Piolax Inc, Kanagawa, Japan) (**Fig. 2b**). The pseudoaneurysm and splenic artery trunk were embolized using NBCA (Histoacryl; B. Braun, Melsungen, Germany) mixed with iodized oil (Lipiodol; Guerbet, Villepinte, France) at a ratio of 1:2 (NBCA: Lipiodol). Postembolization celiac angiography revealed the disappearance of the pseudoaneurysm (**Fig. 2c**). The procedure time from sheath insertion to embolization was 15 min. The 5 F sheath was retained in the right femoral artery in case of rebleeding. After the initial TAE, the patient was admitted to the intensive care unit.

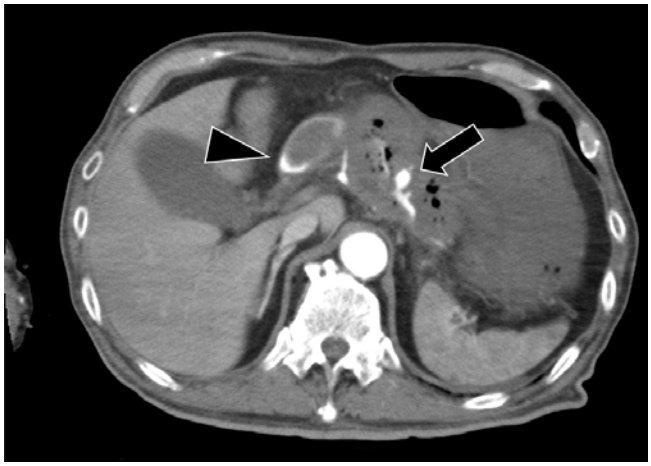

Figure 1. Computed tomography image upon admission
Arterial-phase contrast-enhanced computed tomography showing a ruptured pseudoaneurysm (arrow) of the splenic artery in the pancreatic tumor. Contrast media extravasation is also observed in the gastrointestinal tract (arrowhead). Supplementary Video 1 shows the sequential computed tomography images.

Hematemesis and vital signs that were consistent with shock (systolic blood pressure ≤ 40 mmHg) were observed again 13 h later. Considering the patient was in a serious condition with signs of shock, despite the administration of a blood transfusion using a syringe infusion pump, a second TAE was performed immediately after the arrival of the on-call interventional radiologist without undergoing repeat CECT (**Fig. 3**). The 5 F sheath that was inserted during the first TAE was utilized. Celiac angiography with a 5 F shepherd hook catheter (Hanako Medical Co. Ltd.) displayed massive bleeding from the ostial splenic artery, although blood flow in the previously treated pseudoaneurysm remained occluded (**Fig. 3a**). The splenic artery was catheterized with a 2.0 F tip microcatheter (Bobsled Allorounder; Piolax Inc.) and a 0.014-inch steerable guidewire (Labyrinth Noah; Piolax Inc.) (**Fig. 3b**). The splenic artery was embolized using NBCA mixed with Lipiodol (Guerbet) at a ra-

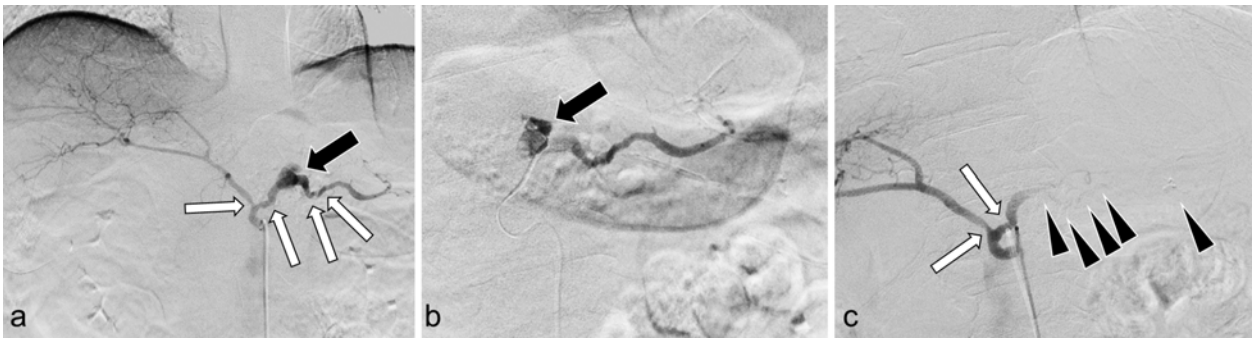

Figure 2. First transcatheter arterial embolization
(a) Celiac arteriography shows a pseudoaneurysm (black arrow) of the splenic artery. White arrows indicate caliber irregularities of the splenic and common hepatic arteries.
(b) A pseudoaneurysm (black arrow) was selected using a microcatheter. Using an NBCA and lipiodol mixture, the pseudoaneurysm and the main trunk of the splenic artery were embolized.
(c) Celiac arteriography after embolization shows the disappearance of the pseudoaneurysm and occlusion of the splenic artery. Arrowheads indicate the distribution of casts in the splenic artery. Caliber irregularities of the proximal splenic and common hepatic arteries remained (white arrows).

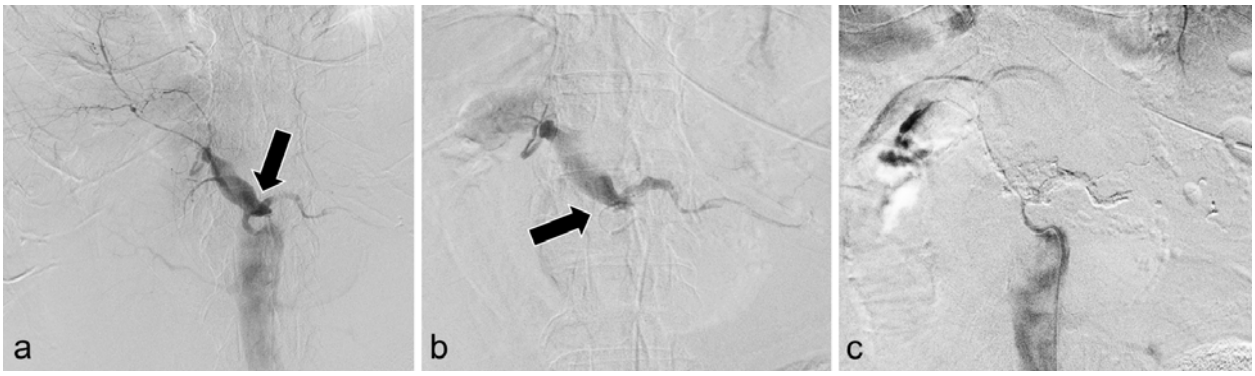

Figure 3. Second transcatheter arterial embolization
(a) Celiac arteriography shows extravasation of contrast medium from the root of the splenic artery (arrow). The pseudoaneurysm that was treated with the first TAE remains occluded.
(b) The splenic artery was selected using a microcatheter (arrow). The splenic artery was embolized using an NBCA and lipiodol mixture. The celiac and hepatic arteries were unintentionally embolized.
(c) Celiac arteriography after embolization shows the disappearance of active hemorrhage; nevertheless, the branch vessels of the celiac artery are not shown.

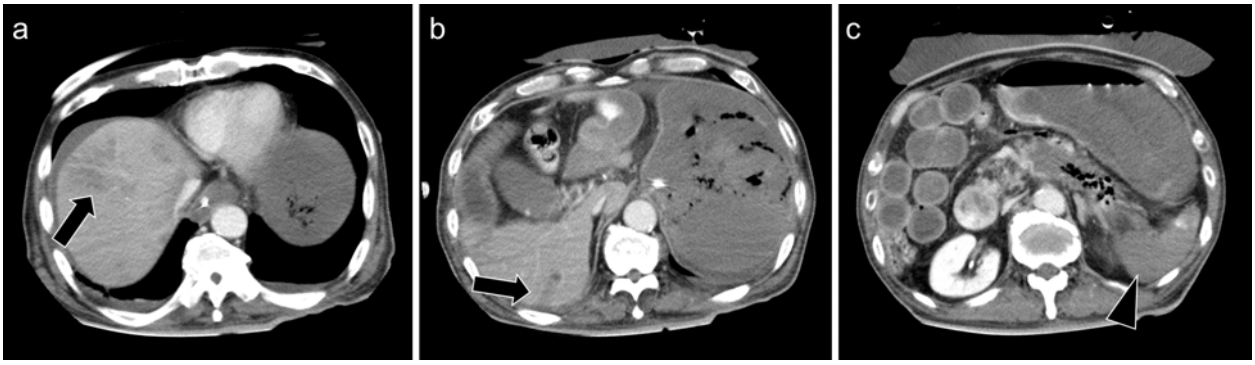

Figure 4. Computed tomography images immediately after the second transcatheter arterial embolization
(a, b, c) Venous-phase contrast-enhanced computed tomography shows partial infarction of the liver (arrows) and entire infarction of the spleen (arrowhead).

tio of 1:2 (NBCA: Lipiodol), and hemostasis was attained. However, the celiac and common hepatic arteries were unintentionally embolized (**Fig. 3c**). The procedure time for the second TAE was 14 min.

CECT immediately after the second TAE showed no active bleeding or pseudoaneurysm; however, splenic and hepatic infarcts developed (**Fig. 4**). Splenic abscesses, pancreatitis, and gastrointestinal necrosis were absent. The patient's condition improved with conservative treatment, and he was discharged from our hospital without rebleeding 25 days after arrival. However, radical treatment of pancreatic cancer was challenging, and the patient was transferred to another hospital for palliative care.

Discussion

We present a case of a patient with invasive pancreatic carcinoma involving most of the splenic artery who developed life-threatening bleeding that was treated by repeated TAE using NBCA. Although embolization is performed for a pseudoaneurysm, it can cause rebleeding shortly afterward if the embolization is not performed appropriately [3]. In the present case, the tumor had invaded most of the splenic artery; hence, both the pseudoaneurysm and splenic artery with irregularity should initially have been treated as a sequential lesion. The reasons for the rebleeding in the present case are predicted to be as follows: (1) Embolization increased splenic arterial pressure more proximal to the embolized portion, and (2) the pancreatic tumor and associated pancreatitis involved the entire splenic artery, including the ostial site. Elevated arterial pressure may prompt splenic arterial rupture and more active bleeding. Moreover, the splenic artery is extensively involved in pancreatic tumors with fistulas in the gastrointestinal tract, resulting in massive rebleeding [3, 5]. We posit that prophylactic embolization of the splenic artery, including its root, should be considered in the first TAE, which might be potentially useful for preventing rebleeding. In other words, after rapid hemostasis was obtained via embolization of the splenic artery using NBCA, embolization using coils at the origin of the splenic artery should have been performed. Furthermore, given that the common hepatic artery showed caliber irregularity, another

treatment option to prevent rebleeding is stent graft placement from the celiac to the common hepatic artery after embolization of the splenic artery.

Embolization with coils or angioplasty with a stent graft from the distal to the proximal aspect of the pseudoaneurysm is ideal for the endovascular treatment of the pseudoaneurysms of the splenic artery [6]. However, performing these treatments in unstable patients who require urgent hemostasis due to massive active bleeding as they take time is challenging. Additionally, navigation of the delivery system and deployment of the stent graft for the commercially available device has a higher risk of iatrogenic vessel injury for the tortuous and fragile target vessel. However, angioplasty with a stent graft is a feasible technique and has the potential merit to preserve the parent artery. Flow control with a balloon catheter may be an alternative technique to achieve transient hemostasis during the coil embolization procedure. Nevertheless, the balloon catheter may require several minutes of test dilatation and air removal to use the catheter safely. Gastroduodenal bleeding due to the rupture of a splenic pseudoaneurysm is rare [7]; however, it causes massive active bleeding due to low internal pressure in the gastrointestinal tract. Invasion of the duodenal wall and splenic artery by the neighboring pancreatic tumor leads to gastrointestinal bleeding through the duodenal fistula. We embolized the ruptured pseudoaneurysms of the splenic artery using only the NBCA and lipiodol mixture in unstable patients. This treatment may be considered in the case of a life-threatening emergency because it provides rapid hemostasis [8, 9]. Furthermore, NBCA provides definitive hemostasis even in patients with coagulopathy [9]. However, it may be a challenging and controversial treatment strategy because of the high risk of nontarget embolization. In the present case, embolization from the splenic to the celiac artery was finally performed, and complications such as splenic and focal hepatic infarctions were treated conservatively. One important limitation of this treatment is the difficulty in predicting the degree of complications. Migration of fragmented NBCA into the peripheral branches to the duodenum, residual pancreas, and gallbladder may cause other serious complications. Additionally, NBCA injection requires operator skill, and if performed by a less experienced operator, the possibility of serious complications is likely to increase. In our case, since the patient was unstable with a systolic blood pressure < 40 mmHg, we used the NBCA and lipiodol mixture for the embolization of a large vessel, namely, the splenic artery. Generally, however, glue embolization of a pseudoaneurysm of a large artery is not recommended. Coil embolization or stent graft placement should be performed as long as the patient's condition is sufficiently stable to allow it.

In this report, we present a case of rebleeding from the root of the splenic artery after a short interval following embolization with an NBCA and lipiodol mixture of a ruptured pseudoaneurysm of the splenic artery. When treating a pseudoaneurysm of the splenic artery extensively affected by a pancreatic tumor and pancreatitis, the entire splenic artery between distal to the pseudoaneurysm and the origin of the splenic artery may also need to be embolized to prevent rebleeding. Additionally, another treatment option to prevent rebleeding may be stent graft placement from the celiac to the common hepatic artery after the embolization of the splenic artery.

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Conflict of Interest: None
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