Case Report

Genuine splenic artery aneurysm rupture treated by N-butyl cyanoacrylate and metallic coils under resuscitative endovascular balloon occlusion of the aorta

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Case: A 66 year-old woman who presented with sudden lower abdominal pain was transferred to our emergency room. Vital signs were stable on arrival at the hospital, but immediately became unstable. Systolic/diastolic blood pressure and heart rate were 66/33 mmHg and 70 b.p.m., respectively. Computed tomography scanning showed splenic artery aneurysm rupture and extravasation. The patient was treated non-operatively and definitively by endovascular therapy comprising resuscitative endovascular occlusion of the aorta for hemodynamic control, N-butyl cyanoacrylate, and metallic coils as an embolization material.

Outcome: On admission day 3, she was enrolled in another department and admission day 54, she was discharged.

Conclusion: Although resuscitative endovascular occlusion of the aorta and N-butyl cyanoacrylate is known to be effective, the use of resuscitative endovascular occlusion of the aorta with transcatheter arterial embolization and N-butyl cyanoacrylate for non-traumatic bleeding has not previously been reported. By combining and adapting these devices, their applications in endovascular management may be increased.

Key words: Non-traumatic, resuscitative endovascular balloon occlusion of the aorta, shock, splenic artery aneurysm, transcatheter arterial embolization

INTRODUCTION

VISCERAL ARTERY ANEURYSMS are rare, but important clinical entities. Rupture of a splenic arterial aneurysm (SAA) is associated with a high mortality rate of up to 36%.¹ Although the majority of SAA cases are asymptomatic, the signs of SAA rupture can vary from abdominal pain or chest pain to cardiovascular collapse.² In emergency cases of SAA with signs, surgical treatment is the best choice; however, for patients considered to be at high risk or inappropriate for surgery, transcatheter arterial embolization (TAE) has recently been proven to be an useful alternative.³ Recently, new devices have emerged for endovascular interventional radiology (IR) procedures and, as a conse-

quence, we could treat emergency patients more safely and definitively. This report describes a case of hemorrhagic shock caused by SAA rupture, which was treated with resuscitative endovascular occlusion of the aorta (REBOA), N-butyl cyanoacrylate (NBCA), and metallic coils.

CASE

A 66-YEAR-OLD WOMAN WHO presented with sudden lower abdominal pain was transferred to our emergency room. Vital signs were stable on arrival at the hospital, but immediately became unstable. Systolic/diastolic blood pressure and heart rate were 66/33 mmHg and 70 b.p.m., respectively, and body temperature was 35.8°C. The patient had an agonized facial expression and a peripheral cold sweat, accompanied by continuous lower abdominal pain. Laboratory test results revealed anemia, thrombocytopenia, and coagulopathy with a hemoglobin concentration of 7.1 g/dL, hematocrit of 20.2%, platelet count of $61 \times 10^3/\mu\text{L}$, and prothrombin activity of 44%. In addition, arterial blood

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Fig. 1. Computed tomography image of the 66 year-old female patient on admission showing extravasation from the splenic artery aneurysm in the intermediate segment of the splenic artery (arrow).

gas analysis showed metabolic acidosis, as indicated by a base excess of -10.2 mmol/L and a lactate level of 6.0 mmol/L. Intravenous lines were secured, and a rapid infusion with normal saline was started. The patient had a history of SAA, and the presence of intra-abdominal free fluid was confirmed with ultrasonography. Therefore, rupture of the SAA was suspected. This was verified by computed tomography scanning, which showed the SAA rupture in the intermediate segment of the splenic artery and extravasation from the artery (Fig. 1). Resuscitative endovascular occlusion of the aorta was initiated by inserting the balloon (Rescue Balloon; Tokai Medical Products, Tokyo, Japan) for hemodynamic maintenance and TAE was carried out. Initially, the balloon was inserted into the right femoral artery and placed above the diaphragm using ultrasound. The balloon was emergently inflated with an increase in systolic/diastolic blood pressure to $164/98$ mm Hg. After recovery from hemorrhagic shock, angiography was initiated. The time from arrival to angiography was 60 min. First, one of two outflow vessels of the SAA was selected with a microcatheter (Estream 2.0; Toray Medical Co., Tokyo, Japan) and coils were indwelled. However, the other outflow vessel could not be selected. Therefore, the SAA was embolized with the injection of 2 mL of 25% NBCA (Histoacryl; B. Braun, Melsungen, Germany) to prevent backflow from the other outflow vessel. The technique of the injection of NBCA was as follows: (i) select proximal portion of the coiled outflow vessel with microcatheter and injected NBCA, (ii) pull back microcatheter to the SAA and inject NBCA continuously until the SAA body is filled, (iii) stop injection of NBCA and withdraw the microcatheter. The balloon was inflated until the

injection of NBCA. After exchanging the microcatheter for a new one, coils were indwelled at the inflow vessel of the SAA, and the procedure was completed by vascular embolization (Fig. 2). During angiography the inflation volume and duration were controlled, and the total occlusion time was 67 min. After treatment, the patient was transferred to the intensive care unit. Ten units of red cell concentrates, 20 units of fresh-frozen plasma, and 10 units of platelet concentrates were administered for 2 days after hospital arrival. Enhanced computed tomography scanning on day 2 of hospital admission showed conclusive embolization of the SAA, the disappearance of extravasation, and colon ischemia at the splenic flexure. We suspected that non-occlusive mesenteric ischemia occurred because of the patient's state of shock on arrival and the REBOA insertion; however, it was conservatively treated. On admission day 3, the patient was enrolled in another department. On admission day 54, she was discharged with ongoing follow-up from the physician.

DISCUSSION

ALTHOUGH SAA REPRESENTS an important clinical entity because of the risk of death if rupture occurs, they are not usually considered in the differential diagnosis of common abdominal complaints. However, SAA is the third most common type of intra-abdominal aneurysm, the most common being abdominal aortic and iliac artery aneurysms.⁴

In general, it is considered that an aneurysm larger than 2 cm is an indication for surgery; an aneurysm between 1 and 2 cm in diameter should be monitored closely with imaging studies every 6 months.¹

Patients presenting with ruptured SAAs are most often treated with splenectomy, removing the portion of the artery containing the aneurysm.⁵ Endovascular IR treatment of SAAs, including coil embolization and stent grafts, has gradually risen in popularity,⁶ and TAE may be carried out for all splenic artery aneurysms.^{7,8} In addition, Dohan *et al.* reported that TAE was an effective and safe treatment option for ruptured visceral artery pseudoaneurysms in hemodynamically unstable patients.⁹ Some investigators also reported that the combination of TAE with NBCA is a safe and effective treatment for visceral pseudoaneurysms, with a high success rate (94%–100%)^{10,11} In the present report, the patient had coagulopathy, and laparotomy was considered to be very high risk. Therefore, TAE was selected for the procedure of hemostasis, with the placement of REBOA to maintain hemodynamics until completion of hemostasis by TAE.

Resuscitative endovascular occlusion of the aorta was invented by Edwards in 1953¹² and was first applied for an abdominal aortic aneurysm.^{13,14} Currently, REBOA is used

Fig. 2. Angiography of a 66-year-old woman with splenic artery aneurysm (SAA) rupture. A, Celiac arteriogram shows contrast extravasation from the SAA in the intermediate segment of the splenic artery. B, Splenic arteriogram after coil embolization of the effluent vessel of the SAA and embolization of the SAA with N-butyl cyanoacrylate–lipiodol mixture. C, After coil embolization of the influent vessel of the SAA, complete hemostasis was achieved.

effectively for achieving emergent hemostasis with traumatic intra-abdominal bleeding;^{15,16} however, there are few reports of non-traumatic cases with REBOA.^{17,18} This is a minimally invasive technique and inserted percutaneously, making it effective for emergent hemostasis whether it is recommended for TAE or surgery. The most prominent feature of REBOA is the ability to rapidly regulate blood flow by balloon inflation or deflation. In the present case, the balloon was inflated and deflated a few times, enabling time to be gained before the fundamental steps of treatment.

N-butyl cyanoacrylate is a non-absorbable liquid ester that polymerizes rapidly in the presence of ionic substances such as blood and saline. When mixed with lipiodol, NBCA becomes radiopaque, and its polymerization is protracted to a timescale of seconds; the polymerization time is inversely proportional to the amount of lipiodol in the mixture.¹⁹ N-butyl cyanoacrylate has two advantages compared with other embolization materials. The primary hemostatic rate of TAE with NBCA is higher than with other devices, specifically, gelatin sponge articles and microcoils, in conditions of coagulopathy.²⁰ In this report, the patient's platelet count was substantially decreased, so normal coagulative function could not be expected. Nevertheless, definitive treatment was achieved with NBCA. The other advantage is the rapid

completion of TAE with NBCA compared with other devices, which is attributed to higher hemostatic rate.²⁰ Disadvantages of TAE with NBCA include its relative expense, instant occlusion of the catheter lumen, difficulty in predicting the precise sites of occlusion, and the steep learning curve for the operator who must determine both the ratio of iodized oil to NBCA, and the volume to be used according to the site and size of the bleeding point. Owing to the disadvantages, the operator must be familiar with using NBCA and pay close attention to use.²⁰

This is the first report of a non-traumatic case treated with the combination of REBOA and TAE with NBCA for intra-abdominal bleeding. Endovascular IR treatment has been considered to be related to the requirement for late surgical conversion in a significant number of patients, and the surgical approach remains the gold standard.²¹ However, advanced endovascular IR treatment using the combination of REBOA and NBCA is capable of definitive treatment.

CONFLICT OF INTEREST

NONE.

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