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Case Report

A ruptured splenic artery aneurysm treated by transcatheter arterial embolization using n-butyl cyanoacrylate–Lipiodol–Iopamidol *,**

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ABSTRACT

N-butyl cyanoacrylate, one of embolic materials, is usually used as a mixture with Lipiodol (N-butyl cyanoacrylate–Lipiodol mixture). N-butyl cyanoacrylate–Lipiodol–Iopamidol was developed by adding a nonionic iodine contrast agent (Iopamiron) to N-butyl cyanoacrylate–Lipiodol mixture. N-butyl cyanoacrylate–Lipiodol–Iopamidol has lower adhesiveness than N-butyl cyanoacrylate–Lipiodol mixture and the ability to form a single large droplet. We report the case of a 63-year-old man with a ruptured splenic artery aneurysm treated by transcatheter arterial embolization using N-butyl cyanoacrylate–Lipiodol–Iopamidol. He was referred to the emergency room because of sudden onset of upper abdominal pain. A diagnosis was established using contrast-enhanced computed tomography and angiography. Emergency transcatheter arterial embolization was performed, and the ruptured splenic artery aneurysm was successfully embolized using a combination of coil framing and N-butyl cyanoacrylate–Lipiodol–Iopamidol packing. This case demonstrates the usefulness of a combination of coil framing and N-butyl cyanoacrylate–Lipiodol–Iopamdol packing for the embolization of aneurysms.

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Introduction

Aneurysms have walls composed of 3 layers: intima, media, and adventitia. The weakness of the blood wall causes the artery dilatation. Hypertension, liver transplantation, cirrhosis, and pregnancy are risk factors of aneurysms [1,2]. In contrast, pseudoaneurysms are abnormal vascular dilatations formed by an injury to the vessel walls, contained in the adventitia of the artery or by the local hematoma surrounding pseudoaneurysms. Pseudoaneurysms have some etiologies, such as inflammation, trauma, infection, and vasculitis [2,3]. Embolization is a useful treatment for aneurysms and pseudoaneurysms.

N-butyl cyanoacrylate (NBCA) is a liquid embolic material that has been widely used for the embolization of active bleeding, pseudoaneurysms, and arteriovenous malformations, including cases with coagulopathy [4–6]. NBCA is usually mixed with Lipiodol (NBCA–Lipiodol mixture; NL), and the polymerization speed can be adjusted by changing the mixing ratio of NBCA and Lipiodol. However, NL has some problems, such as adherence of the catheter to the vessel wall and occlusion of the catheter lumen [7,8].

To resolve these problems, NBCA–Lipiodol–Iopamidol (NLI) was developed by adding a nonionic iodine contrast agent (Iopamiron) to NL. NLI has lower adhesiveness than NL and the ability to form a single large droplet. It was possible to embolize wide-necked aneurysms using a balloon-assisted technique with no gaps in swine, and it has been suggested that NLI could be useful for the treatment of aneurysms [9,10]. However, no clinical cases of aneurysms treated by transcatheter arterial embolization (TAE) using NLI have been reported. We present a case of a ruptured splenic artery aneurysm (SAA) treated by TAE using NLI.

Case report

A 63-year-old man was transported to the emergency room of a hospital because of sudden onset of upper abdominal pain. He had a history of Parkinson's disease. His blood pressure was 120/80 mm Hg and heart rate, 80 beats/min, and laboratory investigations showed anemia (hemoglobin level, 11.6 g/dL). He was diagnosed with a ruptured SAA ($26 \times 30 \times 30$ mm) with bleb formation and a retroperitoneal and intraabdominal hematoma on contrast-enhanced computed tomography (CT) (Fig. 1). He was transferred to our hospital for treatment on the second day of hospitalization.

In our hospital, non-contrast CT showed that the intraabdominal hematoma had increased in size. Furthermore, progression of anemia was observed (hemoglobin level, 9.1 g/dL). Although his blood pressure was 100/60 mm Hg and stable, emergency TAE was performed.

After placement of a 5 French (Fr) introducer sheath in the right femoral artery under local anesthesia, celiac and splenic artery angiogram confirmed the SAA with bleb formation in the distal splenic artery (splenic hilum area), but extravasation was not observed. The SAA had 3 distal branches that



Fig. 1 – Contrast-enhanced computed tomography images obtained at the previous hospital. (A) An axial image showing hematoma (white arrows) and the splenic artery aneurysm (SAA, white open arrow). (B) An oblique coronal maximum intensity projection reconstruction image showing bleb formation of the SAA (white open arrow).

branched near the proximal neck, and 1 branch had a small aneurysm (Fig. 2A). We attempted to isolate the aneurysm by embolizing all distal branches. However, this was unsuccessful, because the microcatheter could not be advanced into a thin distal branch. Therefore, we changed our treatment strategy and planned to embolize the SAA by coil framing and NLI packing, because a suitable size and volume of coils for tight aneurysm packing could not be prepared as it was an emergency TAE, and embolized 1 distal branch of the SAA, in which a microcatheter could be inserted, by coils to prevent leakage of NLI. Through a 4 Fr catheter (SH-K, MEDIKIT, Tokyo, Japan) placed in the splenic artery, a 2.2 Fr microcatheter (Progreat β 3, Terumo, Tokyo, Japan) was advanced into the distal branch with the small aneurysm, and the branch was embolized using a Target XXL-360 6-mm imes 20-cm coil (Stryker, Kalamazoo, MI). The small aneurysm, less than 2 cm in diameter, was not embolized to reduce the procedure time. To embolize the SAA, first, framing was performed using 4 Target XXL-360



Fig. 2 – Angiograms at embolization of the splenic artery aneurysm (SAA). (A) A splenic artery (SA) angiogram showing the SAA with bleb formation on the left side. Three distal branches of the SAA are confirmed (white arrows) and 1 branch has a small aneurysm (thin white arrow). (B) Fluoroscopic image after embolization of 1 distal branch and coil framing of the SAA. (C-F) Fluoroscopic images of 4 NLI injections, (c) first, (d) second, (e) third, and (f) fourth, show increasing radiopaque area of injected NLI. NLI injections were started far from the proximal neck of the SAA and administered where the NLI was not distributed. Final NLI injection was performed from the proximal vicinity of the SAA. The distal branches (F, white arrows) and the proximal neck (F, white thin arrow) of the SAA were embolized with NLI. (G) Fluoroscopic image after embolization of the proximal neck of the SAA with multiple coils (white arrow). (H) An SA angiogram after postembolization of the SAA showing no SAA filling and blood flow in the distal branch of the SAA maintained by collateral circulation.



Fig. 3 – (A) An oblique coronal maximum intensity projection of contrast-enhanced magnetic resonance imaging performed 1 month after embolization of the splenic artery aneurysm (SAA), and showing the SAA as a low signal intensity area and no blood flow in the SAA (white open arrow). The enhancement of the splenic artery (white arrow) and a distal branch with a small aneurysm (thin white arrow) is maintained. (B) An abdominal radiograph 4 months after embolization of the SAA showing a radiopaque area of NLI in the coil framing and roots of the distal branches of the SAA.

coils (diameter of 24 mm and length of 50 cm) (Fig. 2B), and then NLI was injected into the space between the framed coils in the SAA. NLI was prepared using a nonionic iodine contrast agent, which contained 300 mg of iodine/mL (Iohexol, Fuji Pharma, Toyama, Japan), instead of Iopamiron. The first NLI injection was administered far from the proximal neck of the SAA (Fig. 2C). The second and subsequent NLI injections were administered in the SAA where NLI was not distributed (Fig. 2D and E). Finally, NLI was injected from the proximal vicinity of the SAA until a part of the injected NLI extended into the roots of the distal branches and proximal neck (Fig. 2F). No leakage of NLI distal to the SAA was observed, although flow control of the splenic artery was not performed. NLI injections were performed 4 times, and 3 microcatheters, 2.2 Fr Progreatβ3, 2.6 Fr Carnelian HF-S (Tokai Medical Products, Aichi, Japan), and 2.6 Fr Masters HF (ASAHI INTECC, Aichi, Japan), were used because of occlusion of the catheter lumen after NLI injection. A 3-mL syringe (MEDALLION, Merit Medical, Salt Lake City, UT) was used for NLI injection, and NLI was injected through all 3 microcatheters. After 4 injections of NLI, the proximal artery of the SAA was embolized using a vortex coil (diameter of 5 mm and length of 5.5 cm; Boston Scientific, Fremont, CA) and 3 C-stopper coils (diameter/length were 2 mm/60 mm, 3 mm/100 mm, and 3 mm/40 mm; PIOLAX, Yokohama, Japan) (Fig. 2G). A postembolization angiogram of the celiac and splenic arteries revealed no SAA filling. Blood flow in the distal branch of the SAA was maintained by collateral circulation (Fig. 2H).

Contrast-enhanced magnetic resonance imaging (MRI) was performed 5 days after TAE and revealed no blood flow in the embolized SAA. Although minor splenic infarction was caused by TAE, enhancement in most parts of the splenic parenchyma was maintained by collateral circulation. Anemia was also improved by red blood cell transfusion. The patient was discharged 2 weeks after TAE.

Contrast-enhanced MRI was performed 1 month after TAE, and no blood flow was observed in the embolized SAA (Fig. 3A). Furthermore, abdominal radiography performed 4 months after TAE showed NLI filling in the framed coils and roots of the distal branches of the SAA (Fig. 3B).

Discussion

Several modalities are used for the diagnosis of aneurysms and pseudoaneurysms. Ultrasound (US) and contrastenhanced CT are noninvasive and commonly used. Pseudoaneurysms are depicted as anechoic cystic-like lesions on B-mode US. On Doppler US, a swirling pattern inside the lesions, known as a "yin-yang" sign, is observed [11,12]. On CT imaging, peripheral calcification and mural thrombosis, reflecting pathologic changes, may present in aneurysms [2]. Pseudoaneurysms are enhanced equally to the adjacent main arteries in both arterial and venous phases and maintain their shape on delayed phase; however, the shape of enhancement changes in extravasation [3,12].

The techniques of aneurysm embolization include isolation, packing, and a combination of these 2 techniques. Multiple embolic agents, such as gelatin sponges, metallic coils, NBCA, polyvinyl alcohol, and microspheres, are used for TAE. Metallic coils and liquid embolic material (NBCA) are commonly used for the packing embolization technique [13]. In our case, the ruptured SAA was embolized by the packing technique using a combination of coil framing and NBCA (NLI) injection. To our knowledge, this is the first report of treatment of an aneurysm with NLI in clinical practice.

The advantages of the packing technique using metallic coils, particularly detachable coils, are that it can reduce the risk of migration of metallic coils from aneurysms and make the embolization area easier to control. Although it has been reported that a coil packing density (volume embolization ratio; VER) of 24%-25% is related to lower recanalization of embolized aneurysms [14–16], many coils are necessary to obtain a VER of 24%-25%, and coil packing embolization is expensive. In this case, the SAA was large; therefore, numerous coils would have been necessary, increasing the cost for coil packing embolization. Thus, we used 4 detachable coils for coil framing, and 1 detachable coil and 4 pushable coils for embolization of the distal and proximal branches. Embolization using NLI helped to reduce the number of coils and was considered a very useful method from a medical economic point of view. Furthermore, we believe that the use of NLI contributed to the successful embolization of the SAA because even the largest diameter coils (Target XXL-360 24 mm \times 50 cm) that we could prepare were undersized and not suitable for coil packing of the SAA in this case.

NBCA is a liquid embolic material that is often used as a mixture with Lipiodol. It has been reported that NL is similar in nature to Lipiodol, that is, it is a mass of oil droplet in blood [17–19]; therefore, it is difficult to make a large single droplet and fill the lumen of a large aneurysm using NL in blood flow. However, when trying to create a large droplet under flow control, there is a risk of adhesion of NL to the catheter because of the longer injection time. A mixture of NBCA, Lipiodol, and ethanol (NLE) has been reported to have lower adhesion than NL, is able to form a single large droplet, and is suitable for packing aneurysms [18–20]. However, ethanol is irritating and carries the risk of adverse events. Iopamiron causes less irritation than ethanol; therefore, NLI is considered to have a lower risk of irritative adverse events than NLE [9,10].

The balloon-assisted technique or coil embolization of the distal and proximal branches has been combined with NLI or NLE for aneurysm embolization [9,10,18,20,21]. Assisted techniques for embolization (eg, balloon neck plasty and coiling of branches) are considered necessary in embolization with NLI to prevent leakage from aneurysms and retain embolic materials in aneurysms because of the lower adhesion of NLI than that of NL. In this case, coil framing was used as an assisted technique for embolization. During NLI injection, no leakage of NLI from the SAA was observed, although no flow control of the splenic artery was performed. NLI injected into the SAA was shown to remain in the SAA on abdominal radiography 4 months after TAE. This indicates that coil framing is useful to prevent leakage of NLI from aneurysms and retain NLI in aneurysms, similar to the balloon-assisted technique or coil embolization of the distal and proximal branches.

In previous reports, a 1-mL syringe (MEDALLION) and 2.1 or 1.9 Fr microcatheters were used for injection of NLI [9,10]. In this case, a 3-mL syringe (MEDALLION) and 3 microcatheters, 2.2 Fr Progreat β 3, 2.6 Fr Carnelian HF-S, and a 2.6 Fr Masters HF, were used. We could inject NLI, but felt high resistance during injection, although the microcatheters had a wider catheter lumen than those in previous reports. A 1-mL syringe (MEDAL-LION) is recommended for injection of NLI, even when using a microcatheter with a wide catheter lumen.

Although Iohexol, instead of Iopamiron, was used for preparing NLI in this case, obtaining a single large droplet was still possible. The use of nonionic iodine contrast agents other than Iopamiron in NLI has not been reported. This report indicates that using nonionic iodine contrast agents other than Iopamiron in NLI is feasible.

Conclusion

In conclusion, NLI was useful for packing a ruptured large SAA, and combining NLI packing with coil framing would be useful in preventing NLI leakage from aneurysms and retaining NLI in aneurysms. In addition, using nonionic iodine contrast agents other than Iopamiron in NLI is feasible.

Authors' contributions

All authors contributed to data analysis and drafting or revising the article, gave final approval for the version to be published, and agreed to be accountable for all aspects of the work.

Patient consent

Informed consent was obtained from the patient included in this study.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2023.04.005.

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