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Case Report

Retrieval of N-Butyl-2-Cyanoacrylate Glue Migrated to the Vertebral Artery via Dangerous Anastomosis, Using the Stent-Retriever Aspiration Technique, during Dural Arteriovenous Fistula Embolization: A Case Report

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Objective: We report a case of accidental N-butyl-2-cyanoacrylate (NBCA) glue migration into the vertebral artery (VA) via dangerous anastomosis during transarterial embolization (TAE) for transverse sinus (TS)-dural arteriovenous fistula (DAVF), which was rescued by mechanical retrieval using a stent retriever and aspiration devices.

Case Presentation: A 49-year-old right-handed female patient was admitted to our hospital with motor aphasia. MRI revealed congestion in the left temporal and occipital lobes, involving a small hemorrhage. DSA revealed a DAVF complicated by a sinus thrombus in the left TS. The DAVF was mostly fed by the left occipital artery (OA) and drained into the cortical veins of the temporal and occipital lobes through the patent part of the sinus. TAE was performed via the left OA with low-concentration NBCA. However, NBCA glue migrated into the left VA through a dangerous anastomosis, and a left VA angiogram revealed severe VA stenosis and floating NBCA glue. There was a fragile attachment of the NBCA glue to the arterial inner wall; therefore, we successfully retrieved the NBCA glue with a stent retriever and aspiration devices without complications. Finally, TAE was performed using another feeder, and the DAVF was completely obliterated.

Conclusion: TAE using NBCA is useful for the treatment of DAVF; however, it should be noted that there is a risk of migration via potential anastomotic routes. Low-concentration NBCA glue can be retrieved using these devices in limited cases.

Keywords N-butyl-2-cyanoacrylate glue, dural arteriovenous fistula, transarterial embolization, migration, mechanical retrieval

Introduction

A dural arteriovenous fistula (DAVF) is an arteriovenous shunt located in any dural region, most commonly in the transverse-sigmoid sinus (TSS).[1,2] A DAVF is usually fed by several dural branches of the external carotid artery (ECA) or vertebral artery (VA) and drained into the sinus and cortical veins. A DAVF with cortical venous reflux is at high risk for hemorrhage or nonhemorrhagic neurological deficits, and requires aggressive treatment.[2,3] Transarterial embolization (TAE) using N-butyl-2-cyanoacrylate (NBCA) has been accepted as an effective treatment for DAVF.[4] However, NBCA-related complications could occur during endovascular embolization, including NBCA glue migration, occlusion of normal territories, and catheter adhesion to the vessel wall due to its fast-acting adhesiveness.[4,5] We report a case of an accidental NBCA glue migration into the VA via dangerous anastomosis during TAE for transverse sinus (TS)-DAVF, which was rescued by mechanical retrieval using Trevo and Penumbra devices.

Case Presentation

A 49-year-old, right-handed female patient with motor aphasia was admitted to our hospital. MRI revealed congestion in the left temporal and occipital lobes, with a small

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hemorrhage (**Fig. 1A** and **1B**). DSA revealed that the DAVF was shunted around the middle part of the left TS, which was mostly fed by the left occipital artery (OA). The left TS was occluded; however, the fistula drained into the cortical veins of the temporal and occipital lobes through the patent part of the sinus (**Fig. 1C**–**1F**). Transvenous embolization was initially attempted, but the microcatheter could not reach the fistula. Therefore, TAE was performed via the mastoid branch of the left OA, which was the main feeder artery, under general anesthesia. When the microcatheter was advanced into the distal part of the mastoid branch, a catheter-induced spasm occurred. After waiting period, a nicardipine injection was administered, but no improvement was observed. The proximal part of the mastoid branch, with a part of OA, was reluctantly packed with platinum coils and 20% NBCA. Although it was a proximal occlusion without the glue penetration into the shunt point, angiography after embolization showed disappearance of the DAVF (**Fig. 2A**), and the patient's symptom improved.

An eight-day follow-up angiography showed a residual shunt (**Fig. 2B**), and an additional TAE was performed. An ECA angiography revealed that the DAVF was fed by the muscle branch of the OA and the posterior convexity branch of the left middle meningeal artery (MMA), which were not visualized during the first treatment. The left VA angiogram showed no feeder artery to the DAVF. A 6-French (Fr) guiding catheter (Fubuki; Asahi Intecc, Aichi, Japan) was placed in the left ECA under general anesthesia. A coaxial microcatheter system, a 4.2-Fr intermediate catheter (Fubuki), and a microcatheter (Headway 17; Terumo, Tokyo, Japan) were used to treat the OA. Selective angiography of the OA muscle branch revealed that the muscle branch was connected to the OA distal mastoid branch, which was occluded with the first treatment (**Fig. 2C**). Our treatment plans were occlusion of the muscle branch and embolization of DAVF via MMA with NBCA. The microcatheter was wedged into the muscle branch, and embolization with 20% NBCA was performed under biplane blank roadmap. After embolization, it was observed that part of the NBCA glue migrated into the left VA via muscle branch anastomosis (**Fig. 3A**–**3C**). A left VA angiogram revealed severe VA stenosis and floating NBCA glue (**Fig. 3D**). It was a potential risk for critical infarction, so a decision was made to retrieve the NBCA glue using mechanical thrombectomy devices. An 8-Fr balloon-guiding catheter (FlowGate^2; Stryker, Kalamazoo, MI, USA) placed in the left VA via an 8-Fr femoral sheath. For the initial retrieval attempt, a direct aspiration first pass technique, which is less invasive, was used. An aspiration catheter (Penumbra ACE68; Penumbra, Alameda, CA, USA) was advanced proximally to the NBCA glue, and the Penumbra ACE68 was withdrawn under the continuous aspiration using the penumbra pump. However, a subsequent angiogram revealed retrieval failure. The second pass attempt was performed using the stent-retriever aspiration technique. The microcatheter (Marksman; Medtronic, Minneapolis, MN, USA) was advanced through the NBCA glue over the microguidewire (Chikai; Asahi Intecc), and subsequently a 4 × 30 mm Trevo (Stryker) was led and deployed into an acceptable position. The balloon of the guiding catheter was inflated and the Trevo and Penumbra devices were carefully withdrawn under continuous aspiration (**Fig. 4A**). The NBCA glue was removed, but the NBCA glue got trapped at the tip of the guiding catheter outlet due to its large size. Therefore, the guiding catheter was pulled out slowly maintaining continuous aspiration using a 50-cc syringe. Thus, the NBCA glue was successfully retrieved, and an after-retrieval angiogram revealed VA recanalization without slow flow and distal embolization (**Fig. 4B** and **4C**). Finally, the posterior convexity branch of the left MMA was occluded using 20% NBCA. The angiography following the second TAE showed disappearance of the DAVF. Additionally post-intervention MRI showed no signs of intracranial hemorrhage and infarction (**Fig. 5**). A nine-day follow-up angiography showed complete obliteration.

Discussion

Here, the DAVF was an uncommon condition because the left TS was occluded. However, the shunting portion was the middle portion of the TS and drained into the cortical veins via the patent part of the sinus. We hypothesized that the development of DAVF was related to sinus thrombosis, which led to the opening of small physiological arteriovenous pathways in the wall of the sinus or abnormal connections formed during recanalization.[6] TAE is an accepted therapeutic alternative for DAVF, such as the isolated sinus type, when the transvenous approach is not feasible. After the first treatment, the DAVF was fed by the OA muscle branch via the OA mastoid branch and MMA branches. So, our second treatment plans were occlusion of the OA muscle branch and embolization of the DAVF via MMA with NBCA. There could have been networks between the muscle branches and the OA mastoid branch; thus, an embolization of the muscle branch with low-concentration NBCA nearest to the mastoid branch was performed. Retrospectively, the wedged injection with the low-concentration NBCA from the muscle branch of OA was misjudged because there was dangerous anastomosis between OA and

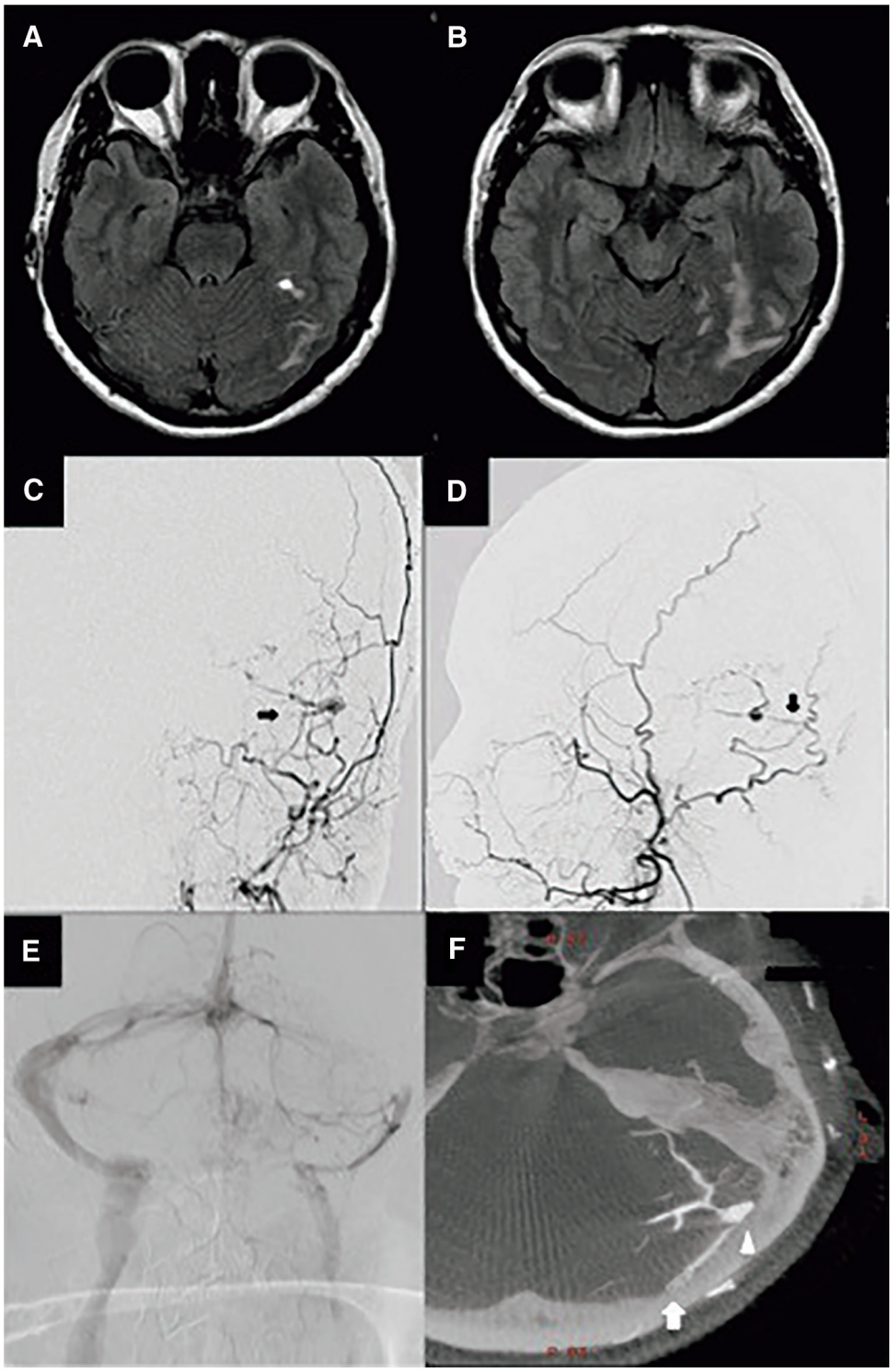

Fig. 1 A 49-year-old, right-handed female presented to our hospital with motor aphasia. (**A** and **B**) Images obtained with MRI–FLAIR showing high-intensity area in the left temporal and occipital lobes involving a small hemorrhage. (**C** and **D**) Anterior-posterior and lateral views of left ECA angiogram showing the arteriovenous fistula shunting at the left TS (black arrow), which is fed by OA mastoid branch and drained into the cortical veins retrogradely. (**E**) Anterior–posterior view of left VA angiogram showing the left TS occlusion. (**F**) Multiplanar reformation images of con-beamed CT obtained from left ECA 3D rotational angiography showing the shunting area at the middle part of the transvers sinus (white arrow) and draining into the cortical veins retrogradely via lateral tentorial sinus (white arrowhead). ECA: external carotid artery; OA: occipital artery; TS: transverse sinus; VA: vertebral artery

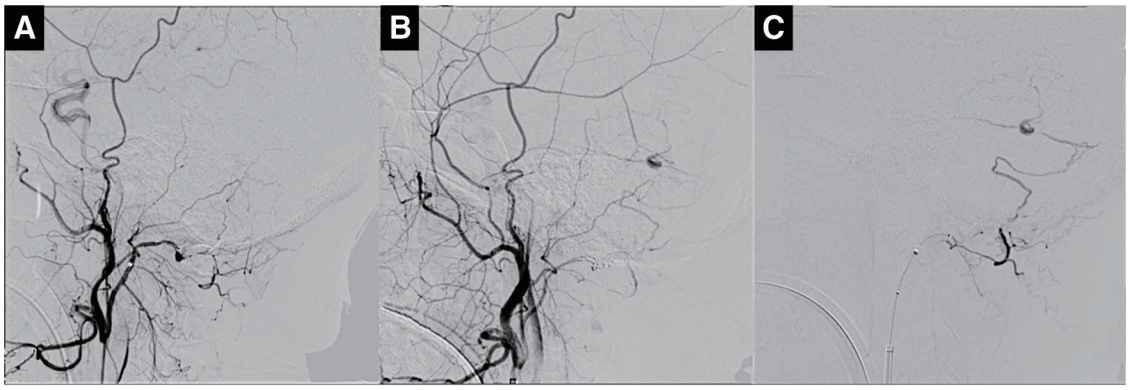

Fig. 2 (**A**) Lateral view of the left ECA posttreatment angiogram showing partial occlusion of OA and the DAVF disappearance. (**B**) Lateral view of the left ECA follow-up angiogram showing residual shunt, which was fed by the OA muscle branch and posterior convexity of the left MMA. (**C**) Lateral view of selective angiogram of the OA muscle branch showing the connection with the OA distal mastoid branch, which was occluded with the first treatment. DAVF: dural arteriovenous fistula; ECA: external carotid artery; MMA: middle meningeal artery; OA: occipital artery

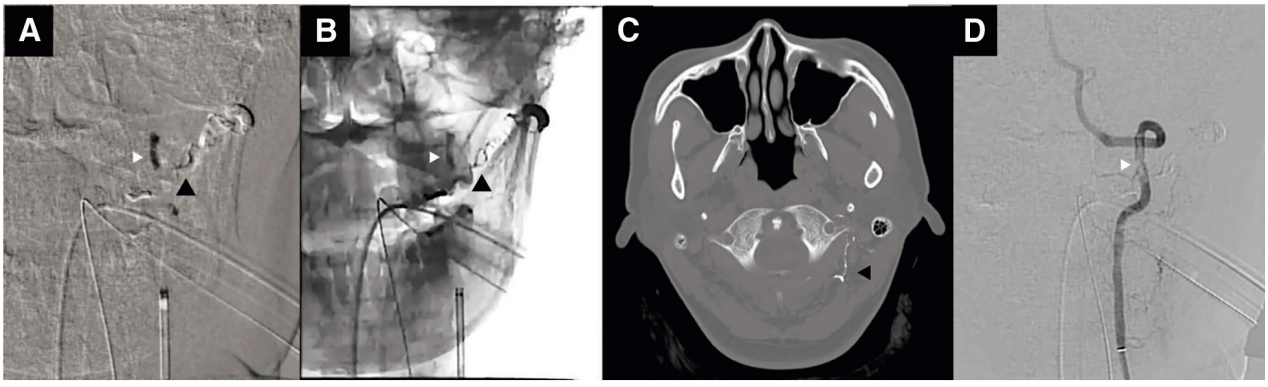

Fig. 3 (**A** and **B**) Anterior–posterior view of blank roadmap and fluoroscopic images showing the migrated NBCA glue (white arrowheads) to VA through the OA muscle branch (black arrowheads) at the C1 level. (**C**) Postembolization CT showing the OA muscle branch (black arrowhead) occluded by NBCA glue at the C1 level. (**D**) Anterior–posterior view of left VA angiogram showing severe stenosis and the migrated NBCA glue (white arrowhead). NBCA: N-butyl-2-cyanoacrylate; OA: occipital artery; VA: vertebral artery

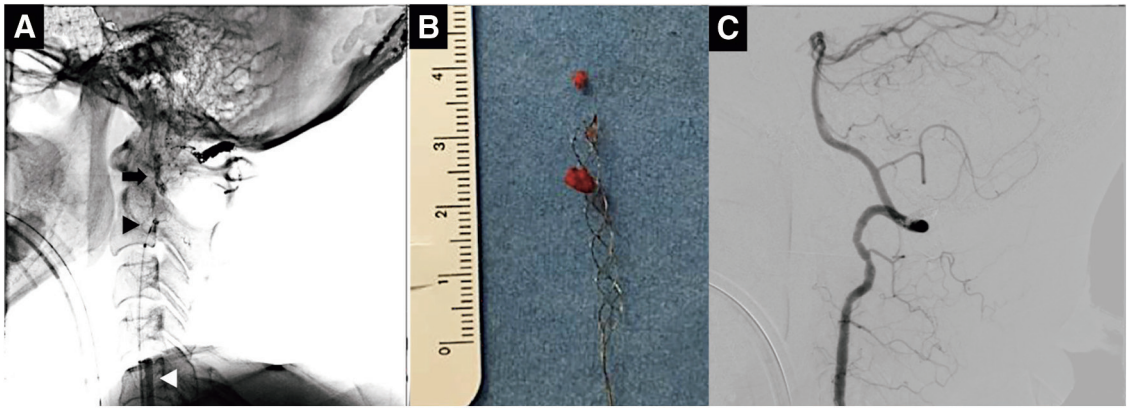

Fig. 4 (**A**) Fluoroscopic image showing retrieval of the migrated NBCA glue using Trevo (black arrow) and Penumbra ACE68 (black arrowhead) under inflation of the balloon-guiding catheter (white arrowhead). (**B**) Image of the NBCA glue caught on the outside of the stent. (**C**) Postretrieval left VA angiogram showing recanalization without stenosis and slow flow. NBCA: N-butyl-2-cyanoacrylate; VA: vertebral artery

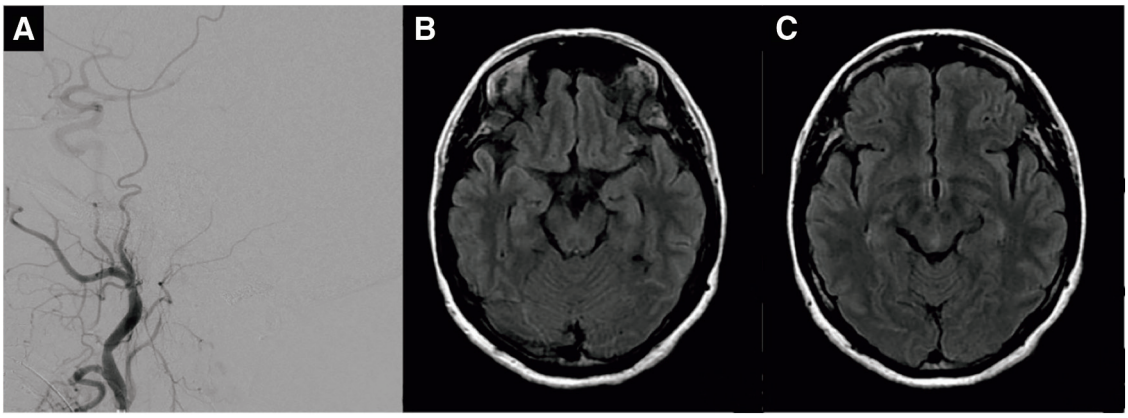

Fig. 5 (**A**) Lateral view of left ECA angiogram showing no DAVF after embolization. (**B** and **C**) Images obtained with MRI–FLAIR showing no abnormal signals after embolization. DAVF: dural arteriovenous fistula; ECA: external carotid artery

VA, although the anastomosis was not visualized on selective microcatheter angiographies. The OA has potential anastomotic routes connected to the VA, which is a remnant of the proatlantal arteries at C1 and C2 levels.[7] The muscle branches, which arise from the horizontal portion of the OA, are the most common routes to the VA. The best imaging condition to inject NBCA is under discussion. Here, NBCA injection was administered under biplane blank roadmap and fluoroscopy; however, the migration of NBCA glue during the procedure was not noticed. There were some reasons for the NBCA glue delayed recognition. During injection to small vessels, it was difficult to recognize NBCA glue because of a small dose, vulnerability to movement, even breathing, and overlapping with bones and teeth. Moreover, an untargeted route such as dangerous anastomosis could have not been predicted. NBCA should have been used with caution considering dose of NBCA, imaging condition, and prediction of flow directions. Occlusion of muscle branch with coils or embolization of DAVF first from MMA feeders could have been alternative methods.

NBCA is a fast-acting and strong adhesive that is widely used to occlude DAVF transarterially with an occlusion rate of 30%–60%.[4,8] In contrast, the reported complications of embolization with NBCA are tissue ischemia, hemorrhage, systemic or local reactions (such as nausea or vomiting, high fever, and regional pain), catheter adhesion to blood vessels, and recurrence.[5,9,10] NBCA glue migration into untargeted vessels is an especially high-risk factor for critical stroke. Once the NBCA glue migration occurs, it is difficult to remove the migrated NBCA glue because of its strong adhesion to the vessel wall. Therefore, NBCA-migrated cases without clinical symptoms are recommended for careful clinical monitoring. However, cases with clinical symptoms and high risk for critical complaint need more aggressive treatment. Here, the migrated and floating NBCA glue posed a risk of distal embolization; therefore, dealing with the complaint was needed. A patent contralateral VA and sufficient collateral posterior circulation blood flow were identified; therefore, VA occlusion with a coil was an option. Parent artery occlusion is effective for glue migration prevention; however, there are potential risks of ischemic events due to hemodynamic change and thrombus of stump vessel. According to the Japanese Registry of Neuroendovascular Therapy 3, the rate of ischemic complications of parent artery occlusion for unruptured VA aneurysms was 18%, which of nearly half cases were transit symptoms.[11] Stent deployment is another option for trapping migrated glue. However, stent-related thrombosis has also risks for the stent occlusion or thromboembolism in the acute phase. According to a systematic review of stenting for symptomatic extracranial VA stenosis by Stayman et al., the rate of periprocedural ischemic complications is 1.9%.[12] Moreover, Coelho et al. reported that the incidence rate of acute carotid stent thrombosis was 0.36%–33%; furthermore, the emergency and antiplatelet noncompliance were risk factors.[13] Therefore, these methods require pre- and postprocedural antithrombotic treatment to prevent ischemic events. Open surgery with or without bypass may also be an option; however, this is obviously more invasive and not necessarily more effective. The 2nd and 3rd segments of the VA are extremely difficult to expose as it runs within the bony canal formed by foramen transversarium. Berguer et al. reported the outcome of surgical reconstruction of the 369 consecutive extracranial VAs with the stroke and death rate of 1.9%–5.1%.[14] Hence, it could be worth to remove NBCA glue using mechanical thrombectomy devices. Retrieval cases of migrated materials, such as coil, stent, or nonadhesive agent

Table 1 Summary of the retrieval cases of migrated NBCA glue after trans-arterial embolizaion in literature

Study	Pt age/sex	Disorder	Target vessel	NBCA concentration	Migrated timing	Migrated location	Retrieval devices	Complication
Fahed et al.[18]	29/male	AVM of claustrum	Branch of MCA	20%	On embolizaion	Straight sinus	Solitaire	None
Ahmed et al.[19]	77/female	DAVF of TSS	Branch of OA	25%	On embolizaion	TSS	Snare	None
Chauhan et al.[20]	70s/female	Orbital meningioma	Direct puncture of tumor	50%	7 hour after embolization	MCA	Solitaire and Penumbra	Infarction
Present case	49/female	DAVF of transverse	Branch of OA	20%	On embolizaion	VA	Trevo and Penumbra	None

AVM: arteriovenous malformation; DAVF: dural arteriovenous fistula; MCA: middle cerebral artery; NBCA: N-butyl-2-cyanoacrylate; OA: occipital artery; Pt: patient; TSS: transverse-sigmoid sinus; VA: vertebral artery

(Onyx), using retrieval devices have been reported.[15–17] Moreover, a few recent cases have reported successful retrieval of NBCA glue (**Table 1**). There were two retrieval cases of migrated NBCA glue during TAE for intracranial arteriovenous malformation and DAVF due to venous outflow obstruction using the Solitaire FR or Snare-kit system.[18,19] Chauhan et al. reported a retrieval case of NBCA glue that migrated into the middle cerebral artery 7 hours after TAE for orbital meningioma.[20] They retrieved the NBCA glue using the Solitaire and Penumbra devices.

This is a rare reported case of NBCA glue retrieval that migrated to the VA via dangerous anastomosis using Trevo and Penumbra devices. There was a fragile attachment of the NBCA glue to the arterial inner wall; the use of a stent retrieval was therefore effective in the retrieval of the glue. Furthermore, proximal occlusion with a balloon guiding catheter and continuous aspiration were also effective in avoiding distal embolization. The characteristics of NBCA glue were different from those of a thrombus in the following points. First, the NBCA glue was adhesive to the vessel inner wall, so that retrieval of the NBCA glue could be at risk for vessel damage such as dissection or rupture. Thus, caution had to be exercised during the retrieval of NBCA glue. Second, the NBCA glue was more elastic and harder than a thrombus. The stent could therefore not expand inside the NBCA glue; thus, the NBCA glue was caught on the outside of the stent. The missing NBCA glue could have led to an unexpected embolization with possible disastrous consequences. Finally, low-concentration NBCA glue mixed with lipiodol was visible under fluoroscopy without a contrast agent. A combination technique could be more effective and safer for glue retrieval than a single technique using a stent retriever, aspiration, or snare device. This technique could be an option for the rescue method when low-concentration NBCA glue migrates to the unintentional vessel in limited cases.

Conclusion

A case of successful retrieval of migrated NBCA glue using Trevo and Penumbra devices was reported. TAE using NBCA is useful for the treatment of DAVF; however, it should be noted that there is a risk of migration via potential anastomotic routes. The low-concentration NBCA glue could be retrieved with those devices in limited cases.

Disclosure Statement

The authors report no conflicts of interest concerning the materials or methods used in this study or the findings specified in this paper.

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