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Abstract

Treating carotid blowout syndrome following rupture of giant pseudoaneurysms is difficult because the destroyed parent artery precludes conventional treatment. We present a patient with a ruptured giant pseudoaneurysm that we occluded using a modified internal trapping technique with low-concentration N-butyl-2-cyanoacrylate (NBCA) and a minimum number of coils. An 80-year-old man with a history of chemoradiation therapy for oropharyngeal cancer presented with several episodes of active bleeding from the subsequent tracheostomy site. Radiological examination revealed a giant right common carotid artery (CCA) pseudoaneurysm. Endovascular internal trapping was performed using both NBCA and coils under proximal flow control. We slowly injected 9 ml of low-concentration NBCA, which subsequently filled the entire pseudoaneurysm. We then injected an additional 2 ml of NBCA into the proximal CCA to achieve complete obliteration. No re-bleeding was observed during the 6-month follow-up. Endovascular internal trapping using low-concentration NBCA was feasible to treat a giant CCA pseudoaneurysm. The injected low-concentration NBCA filled the entire pseudoaneurysm without the risk of catheter entrapment.

Keywords

Carotid blowout syndrome, Endovascular embolization, Low-concentration, N-butyl-2-cyanoacrylate, Pseudoaneurysm

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Introduction

Carotid blowout syndrome (CBS) is a devastating complication of surgical and radiation therapies for head and neck cancers. The reported incidence of CBS is 3%–4% for all patients with head and neck cancer, and approximately 40% mortality and 60% severe neurological morbidity were reported when the carotid artery was surgically ligated.[1,2] This syndrome occurs commonly in patients with advanced head and neck cancer with a history of radiation therapy, and the syndrome is also associated with erosion and destruction of the vessel wall.[3] Direct surgical management of CBS is often challenging because exploration and repair in the previously irradiated field are difficult. Thus, endovascular management with either deconstructive permanent occlusion or with reconstructive stent grafting are alternatives to traditional surgical ligation, and these techniques are associated with acceptable outcomes.[4,5] However, in CBS associated with a giant pseudoaneurysm, it is difficult to embolize with coils from distal to the damaged artery to the proximal normal vessel or to deliver a relatively stiff covered stent to traverse the pseudoaneurysm while obtaining adequate distal and proximal seal zones.

N-butyl-2-cyanoacrylate (NBCA) is an adhesive agent that polymerizes in the presence of anions and is widely used as a liquid embolic material. Polymerization time can be prolonged by increasing the ratio of the iodized oil concentration, which allows the NBCA to fully reach and permeate target vessels.[6] To our knowledge, endovascular internal trapping using low-concentration NBCA for a ruptured giant common carotid artery (CCA) pseudoaneurysm has not been reported previously.

We present a patient with a ruptured giant pseudoaneurysm, which we occluded using a modified internal trapping technique with a combination of NBCA and a minimum number of coils.

Case report

An 80-year-old man was referred to the emergency department of our hospital after several episodes of active bleeding from his tracheostomy site. He had a history of right oropharyngeal lateral wall cancer (stage T2N2bM0) 24 years before his presentation. At the time of his diagnosis, he received chemotherapy with cisplatin and fluorouracil followed by conventional radiotherapy with a total dose of 70 Gy to the neck, which resulted in complete remission. Eleven years later, he experienced a minor stroke and underwent bilateral carotid artery stenting for high-grade internal carotid artery stenosis, at another hospital. Twelve years after bilateral carotid artery stenting, he was referred to our cardiovascular department because of a progressive giant subclavian artery (SCA) aneurysm detected on follow-up carotid duplex examination. Supra-aortic three-dimensional computed tomographic angiography (CTA) images showed a right SCA aneurysm (Fig. 1A). Our cardiovascular ream recommended an open surgical approach, but the patient declined direct surgery and preferred minimally invasive treatment. The operation was performed under general anesthesia 9 months prior to his hemorrhagic presentation. First, we performed bypass grafting between the left and right SCAs in an end-to side fashion using an 8-mm ringed Gore-Tex graft (W.L. Gore & Associates, Flagstaff, AZ). Second, two 10-mm-diameter and 3.5-cm long Iliac Extender Endoprostheses (W.L. Gore & Associates) were inserted by overlapping into the right CCA to exclude the origin of the right SCA from an 8-French sheath placed at the CCA just proximal to the carotid stent. Finally, a 14-mm-diameter, 8-mm-long Amplatzer vascular plug (St. Jude Medical Inc., St. Paul, MN) was deployed through a 6-French Destination guiding sheath (Terumo Corp., Tokyo, Japan) into the right SCA to exclude back-flow from the SCA. Follow-up CTA performed 10 days postoperatively confirmed successful exclusion of the SCA aneurysm, with no evidence of leakage (Fig. 1B). Four months after the surgery, he was urgently admitted to the otolaryngology department because of acute dyspnea secondary to vocal cord dysfunction. Axial and reconstructed coronal and sagittal images of neck CT revealed no signs of carotid abnormality. The etiology of vocal cord dysfunction was considered to be late effect of radiation for neck and he underwent emergent tracheostomy. The clinical course after tracheostomy was uneventful and the patient was discharged home. Four months later, he was transferred to the emergency room of our hospital after several episodes of active bleeding from the tracheostomy site. On initial examination, old blood was present in the tracheostomy, but there was no active bleeding. His vital signs were stable, with a hemoglobin level of 8.6 g/l. Supra-aortic CTA showed an irregularly-shaped giant pseudoaneurysm in the right CCA (Fig. 1C). After discussion with the vascular surgeons, we planned to perform endovascular internal trapping using both NBCA and coils.

The endovascular procedure was performed under local anesthesia through the right femoral artery. An 8-French guiding catheter (Neuro EBU; Asahi Intecc Co., Ltd, Aichi, Japan) was advanced into the proximal portion of the stent graft placed in the innominate artery (Fig. 2A). Digital subtraction angiography of the CCA showed a giant pseudoaneurysm associated with arterial disruption (Fig. 2A). To perform the balloon occlusion test (BOT) in the right CCA, we advanced a 6-French balloon guiding catheter (Optimo; Tokai Medical Products, Aichi, Japan) and inflated the balloon in the right CCA. The patient tolerated a 20-minute occlusion test. First, we advanced a microcatheter (Excelsior 1018; Stryker Neuroendovascular, Fremont, CA) into the distal portion of the right CCA. With proximal flow control for the CCA, we placed 13 platinum coils in the distal CCA where the vascular wall was considered normal (Fig. 2B). After the tip of the microcatheter was replaced in the aneurysm, we placed two 24-mm × 50-cm Target XXL coils (Stryker Neuroendovascular) roughly in the aneurysm (Fig. 2B). We prepared a 14% NBCA concentration by diluting the NBCA with lipiodol, then injected the solution slowly and intermittently under fluoroscopic guidance. A total of 9.1 ml of NBCA solution was injected over 4.5 minutes, and we were able to remove the microcatheter without resistance. When injecting the NBCA, we flushed the lumen of the 6-French balloon guiding catheter continuously with 5% dextrose solution to prevent early and abrupt polymerization. We placed a 10-mm × 40-mm Target XXL coil (Stryker Neuroendovascular) in the proximal CCA followed by injecting 2.0 ml of the same concentration NBCA solution. Finally, we occluded the proximal CCA by placing nine platinum coils (Fig. 2C), which achieved complete obliteration of the pseudoaneurysm.

There were no procedure-related complications, and the patient’s postoperative course was uneventful. During the 6-month follow-up, no hemorrhagic event was observed.

Discussion

CBS is classified into three types according to the clinical manifestation, namely acute, impending, and threatened.[1,2,7] Acute CBS is defined as fatal hemorrhage associated with a completely ruptured injured vessel that cannot be controlled with surgical packing alone, and the patient requires immediate resuscitation. Impending CBS describes patients with bleeding episodes that resolve spontaneously or with wound packing. Hemorrhages usually occur through a surgical wound or fistula reaching the esophagus or trachea. Threatened CBS is characterized by exposure of the carotid artery, which is not covered by normal surrounding tissue, and disruption of the arterial wall. A recent meta-analysis of 559 patients with CBS showed that 287 patients (51.3%) presented with acute hemorrhage, 272 patients (48.7%) had threatened and impending presentations, and the involved arteries were the internal carotid artery (23.4%), external carotid artery (39.0%), and CCA (37.2%).[5]

Traditionally, the most common management of CBS is surgical ligation of the carotid artery, despite the high morbidity and mortality rates.[1,2] However, the management paradigm for CBS has shifted from traditional open surgery toward endovascular treatment, which provides acceptable short- and long-term outcomes.[4,5,8,9] According to the National Inpatient Sample from 2003 to 2011, 88.6% of patients underwent embolization procedures, and 11.4% underwent carotid stenting.[4] A recent literature search from 2000 to 2016 identified 559 patients and indicated that 52.8% of the patients underwent coil embolization, 35.2% received covered stent grafts, and 13.4% underwent alternative techniques, including only 13 patients (0.02%) who underwent surgical ligation.[5] The treatment strategy for CBS is determined mainly according to the assessment of the risk of cerebral ischemia from permanent occlusion of the affected carotid artery. A BOT is preferred before CBS treatment; however, this test may not be possible in acute settings where patients require interventional procedures because of unstable vital signs. Furthermore, as many as 15%–20% of patients with negative BOT results who were treated with permanent carotid occlusion developed delayed cerebral ischemia.[10,11]

Endovascular management with permanent occlusion of the carotid artery has been considered effective for treating CBS; however, reconstructive management using covered stents should be considered in patients with risks such as an incomplete circle of Willis or intolerance to BOT. Several studies reported that covered stents may be durable treatment options, with a low risk of perioperative stoke and high technical success rates.[5,12-14] During treatment using covered stents, this relatively stiff device should be delivered to traverse the injured carotid artery site and be deployed across the damaged site with adequate distal and proximal seal zones because inadequate coverage of the pathological lesion is a cause of re-bleeding.[9] In our patient, radiological findings indicated that the severely tortuous CCA distal to the pseudoaneurysm had a degenerated vascular wall, which prevented successful exclusion of the pseudoaneurysm by a covered stent. According to the BOT results, with good collateral flow from the circle of Willis, we excluded covered stent therapy as a possible treatment in our patient.

Permanent vessel occlusion remains the gold standard treatment for CBS, with favorable short and long-term outcomes.[4,5,9] In our patient, a large number of coils was required to completely occlude the giant pseudoaneurysm, which was problematic regarding cost effectiveness. Moreover, delayed coil migration into the airway was a reported rare complication of coil embolization for a pseudoaneurysm in a patient who previously underwent neck irradiation.[15] The possible mechanisms for this complication are explained that coils were placed in aneurysms with more fragile media and intima layers than in true aneurysms, and that irradiated soft tissue around the vessel were not functioning as supportive tissue.[16] Thus, we planned internal trapping with a liquid embolic material using a minimum number of coils, in our patient.

Because of the low viscosity and low tissue toxicity, NBCA is a theoretically optimal permanent embolic material for the treatment of pseudoaneurysms; however, catheter entrapment potentially occurs during injection. For embolization, NBCA is mixed with iodized oil in a ratio from 1:1 to 1:9. An in vitro study showed that the polymerization time increased with the iodized oil ratio, and that polymerization times were similar between ratios of 1:1 and 1:2 and between ratios of 1:3 and 1:5; however, times increased exponentially with ratios of 1:6 to 1:9.[6] Because the time for polymerization is controlled by the mixing ratio, we considered that low-concentration NBCA (1:6 in our patient) would make it possible to permeate the target vessels. We placed two coils approximately in the aneurysm to create a frame inside the aneurysm. This frame was useful to keep the tip of the microcatheter in the center of the giant pseudoaneurysm while injecting the NBCA solution. However, even using low-concentration NBCA, the degree of risk of adhesion of the catheter tip to the vascular wall remains unclear. A recent study reported the usefulness of extremely slow injection of 13% NBCA into the middle meningeal arteries for patients with meningiomas.[17] The reported injection time ranged from 157 seconds to 989 seconds (mean: 364 seconds), and all microcatheters were retracted without difficulty, even if the reflux extended for more than 1cm from the tip of the catheter. Thus, we believe that slowly-injected low-concentration NBCA could permeate the entire space of a giant pseudoaneurysm without the risk of catheter entrapment. Onyx (Medtronic-Covidien, Irvine, CA) is a non-adherent liquid embolic material with favorable permeability and consistency for the treatment of arteriovenous malformations or fistulae. However, Onyx with dimethyl sulfoxide can cause severe inflammation, vasospasm, and endothelial necrosis following intra-arterial injection. Because spontaneous extrusion of Onyx through the scalp was reported in the literature,[18] we believe that Onyx use should be limited in patients with CBS.

Conclusion

Endovascular internal trapping using low-concentration NBCA was feasible for treating a giant CCA pseudoaneurysm when the destructive angioarchitecture prevented conventional access. Low-concentration NBCA could be slowly injected to permeated the entire pseudoaneurysm without the risk of catheter entrapment.

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Declarations of interest

The authors declare that there is no conflict of interest with respect to the research, authorship, and publication of this article.

Ethical statement

Written informed consent was obtained from the patient to publish identifiable information or images within the manuscript.

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Figure captions

Figure 1. Serial three-dimensional computed tomographic angiography (CTA) images of the supra-aortic artery. Preoperative (A) and postoperative (B) CTA images showing successful exclusion of the right subclavian artery aneurysm by inserting a stent graft in the right common carotid artery with crossover bypass grafting between bilateral subclavian arteries. (C) An emergent CTA image obtained during a hemorrhagic event showing a giant pseudoaneurysm in the right common carotid artery.

Figure 2. Right common carotid angiographic images obtained at embolization. (A) Subtracted anteroposterior carotid angiographic image showing a giant pseudoaneurysm associated with arterial disruption. (B) Non-subtracted anteroposterior image showing roughly-packed platinum coils in the pseudoaneurysm. Note the tightly-packed coils in the normal common carotid artery just proximal to the previously placed carotid stent. (C) A postoperative non-subtracted image showing successful internal trapping of the aneurysm by the N-butyl-2-cyanoacrylate and coils. Note the injected N-butyl-2-cyanoacrylate configuration corresponding to the shape of the pseudoaneurysm.


A
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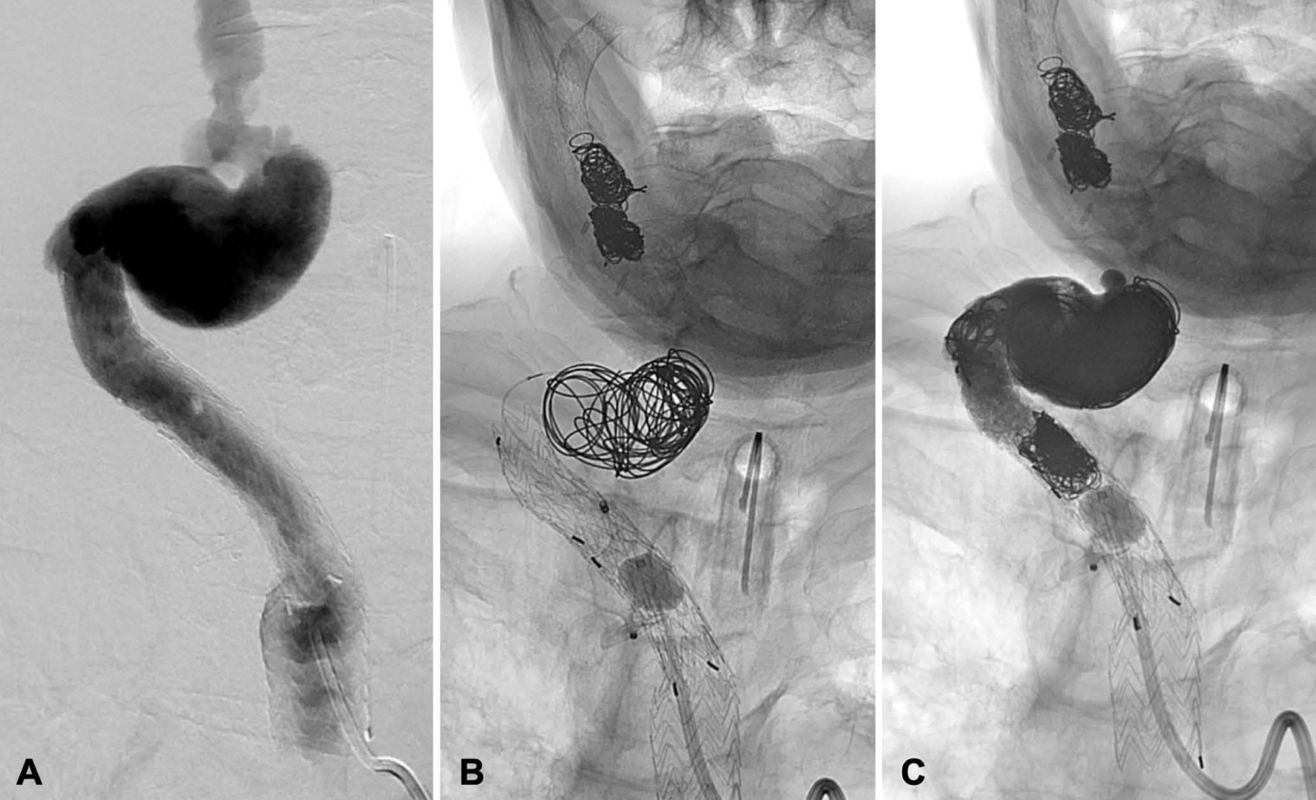
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