

Endovascular Therapy for a Post-irradiated Cervical Pseudoaneurysm at the Carotid Stump: A Case Report

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A nearly 80-year-old man with a history of radiotherapy (RT) and total laryngectomy for laryngeal cancer 25 years previously presented with a rapidly growing mass on the right side of his neck. A huge pseudoaneurysm (pseudoAN) was detected at the right carotid bifurcation, though angiography performed four years previously had shown total occlusion of the internal carotid artery. Stent-assisted coil embolization enabled aneurysm sac shrinkage. Clinicians must be aware that the stump of an arterial occlusion associated with RT can change into a pseudoAN over the long term and must provide follow-up in such cases.

Keywords: radiation-induced, pseudoaneurysm, coil embolization, internal carotid artery, carotid stump

Introduction

Radiotherapy (RT), which is often indicated for the cancer of the head and neck region, can often cause acute or chronic injuries around the radiation site. It is a known characteristic of radiation to cause long-term effects progressing in stages after RT.¹⁾ Scarring, ulceration, and the mucosal fibrosis of the skin, pharynx, and esophagus are well known as late radiation injuries.²⁾ Additionally, sclerotic change of the arterial walls with a reported severity of 20–40% occurs after radiation and gradually worsens.^{3,4)} Meanwhile, pseudoaneurysms (pseudoANs) occurring after RT for laryngeal cancer are rarely observed at the internal carotid artery (ICA) origin or common carotid artery (CCA),^{5,6)} though they are often seen at the petrous portion after RT for nasopharyngeal cancer or skull base tumor.⁷⁾ Here, we report on endovascular therapy for a rare case of secondary aneurysmal change at the residual stump of the post-irradiated cervical ICA occlusion after RT for laryngeal cancer.

Case Report

A nearly 80-year-old man had noticed a rapidly growing mass in the anterior portion of his neck starting three months before presentation. He had a history of radiotherapy (a total

dose of 64Gy) for early-stage laryngeal cancer 25 years previously. A localized recurrence of the cancer three years later had resulted in a total laryngectomy. After the laryngectomy, no cancer recurrence was observed during follow-up. At presentation, the patient was taking no medication for hypertension, ischemic coronary disease, diabetes mellitus, or dyslipidemia.

The patient had suffered from cerebral infarction in the left superior frontal gyrus four years prior to presentation (Fig. 1A). A head and neck angiogram performed at his previous hospital had shown bilateral ICA occlusions and a residual stump at the carotid bifurcation (Figs. 2A and 2B). Right carotid ultrasonography (US) had shown a 1 × 2 cm heterogeneous low echoic mass in the ventral space of the right ICA stump (Fig. 2C). The anterior circulation was supplied through the ipsilateral external carotid artery (ECA) and vertebra-basilar artery via the posterior communicating artery (Figs. 2D–2F). The right A1 segment of the anterior cerebral artery (ACA) was hypoplastic, and the blood flow in the left A2 segment was obviously decreased. N-isopropyl I-123 p-iodoamphetamine (IMP) single photon emission computed tomography (SPECT) had shown a decrease of approximately 10% in cerebral blood flow (CBF) at the left frontal lobe (Fig. 1B). Considering the progressive enlargement of the infarct in the magnetic resonance (MR) diffusion weighted image, it was speculated that hemodynamic stroke of the left A2 territory had occurred when the left ICA severe stenotic lesion was completely occluded.

He was referred to our hospital for further evaluation and treatment of the palpable mass on the right side of the neck below the angle of the mandible. At our institution, the patient was alert and no neurologic deficits were observed. An elastic soft mass approximately 5 cm in diameter was observed in the right lower jaw. An MR angiography (MRA) source image revealed that the mass was a 33-mm lesion at the right anterior portion of the neck that was causing tracheal deviation and compression (Figs. 3A and 3B). From his medical history and the radiological imaging findings, a radiation-induced pseudoAN was suspected. We recognized that urgent therapeutic intervention was needed to prevent aneurysmal rupture. Considering his general condition and advanced age, endovascular treatment was selected.

Endovascular Treatment

Cilostazol (200 mg) and clopidogrel (75 mg) were administered daily starting five days before the procedure.

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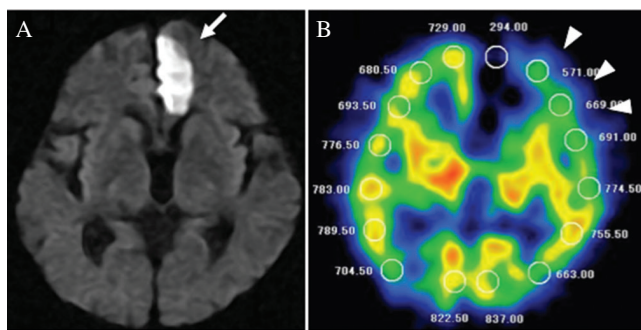


Fig. 1 (A) Diffusion weighted image of the brain obtained four years before the present hospital admission showing acute cerebral infarction in the left superior frontal gyrus (arrow). (B) N-isopropyl I-123 p-iodoamphetamine (IMP) single photon emission computed tomography (SPECT) showing a decrease of approximately 10% in cerebral blood flow at the left frontal lobe (arrowhead).

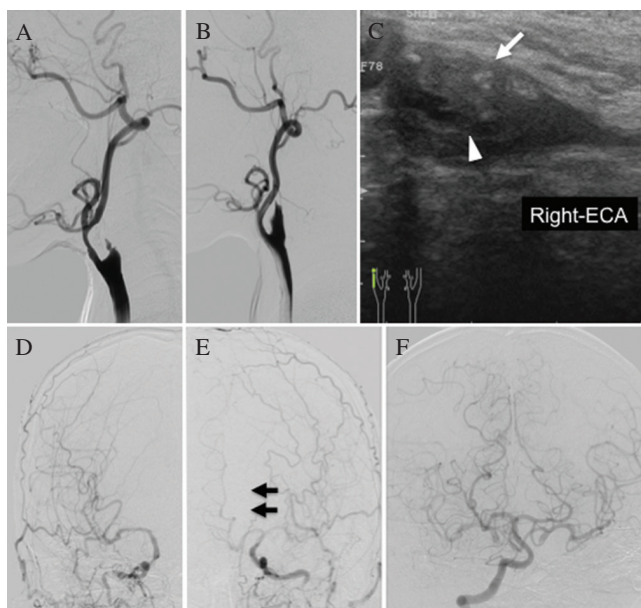


Fig. 2 Right (A) and left (B) common carotid angiograms in the lateral view four years before the present hospital admission showing the residual stump at the bilateral carotid bifurcation. (C) Right carotid ultrasonography showing a 1 × 2 cm heterogeneous low echogenic mass (white arrow) in the ventral space of the right ICA stump (arrowhead) (left side: caudal, right side: cranial). Angiograms of the right common carotid artery (D), left common carotid artery (E), and right vertebral artery (F) in the anterior view showing the collateral flow to the anterior circulation. The right A1 segment of the anterior cerebral artery (ACA) was hypoplastic, and blood flow in the left A2 segment was obviously decreased (E, black arrow).

Stent-assisted coil embolization was attempted under intravenous anesthesia. Through the femoral approach, an Asahi Fubuki™ 8.0 French guiding sheath (Asahi Intecc Co., Ltd., Seto, Aichi, Japan) was introduced into the right common carotid artery. Control angiography revealed that a huge, irregularly shaped aneurysm with a maximum diameter of 27 mm was present at the right carotid bifurcation (Fig. 4A) and that the right ICA origin had been completely occluded as before. A microcatheter (Excelsior™ SL 10,

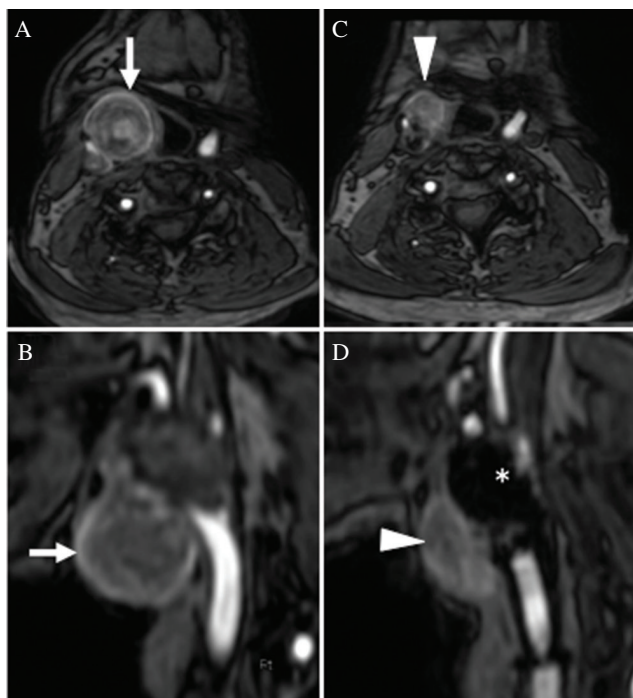


Fig. 3 Preoperative source image in MR angiography at the level of the C5 vertebral body from (A) an axial view and (B) a lateral view showing the 33 mm × 33 mm mass of the thrombosed sac (arrow, point of maximum diameter) and tracheal compression. Follow-up images (C, D) obtained 16 months after the procedure (D, coil mass indicated by asterisk) showing the shrinkage of the mass (18 mm × 18 mm) and disappearance of tracheal deviation (arrowhead).

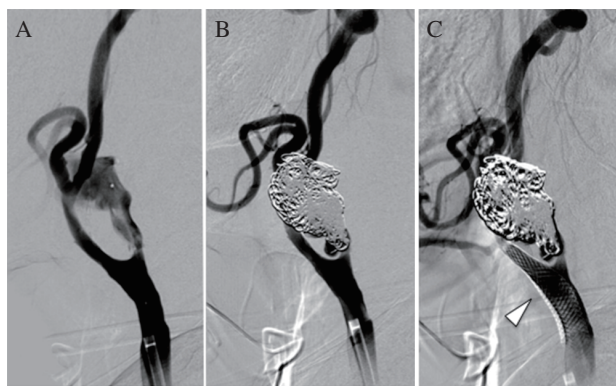


Fig. 4 Preoperative right carotid angiogram in the lateral view (A) showing a huge (14 × 16 × 27 mm) pseudoaneurysm. Postoperative angiogram (B, C) showing the endovascular coiling and stent placement (arrowhead).

Stryker Neurovascular, Fremont, CA, USA) was advanced into the distal portion of the aneurysm. Another microcatheter (Headway 17, MicroVention-Terumo, Tustin, CA, USA) was advanced and positioned at the middle portion of the aneurysm through the same guiding sheath, and a Hydroframe 18™ (14 mm × 45 cm, MicroVention-Terumo) was selected as a framing coil. Tight packing was achieved using hydrogel-coated coils, including HydroSoft™ (MicroVention-Terumo), which were positioned proximal to

the aneurysm for the obliteration of the aneurysmal neck. Although hydrogel-coated coils accounted for approximately 60% of the total length (410 cm) of the coils, residual blood inflow to the aneurysm was still found (Fig. 4B). Therefore, we added stent placement in order to reduce the blood flow to the aneurysm while maintaining the vascular patency of the ECA. Under the distal filter protection consisting of a Spider Fx™ (4.0 mm, Medtronic, Dublin, Ireland) positioned at the trunk of the external carotid artery, a Carotid Wallstent (8 mm × 21 mm, Boston Scientific, Marlborough, MA, USA) was placed at the neck of the aneurysm (Fig. 4C). The post-operative course went well, and the patient was discharged eight days later with no neurological deficits. Sixteen months later, a new MRA source image revealed aneurysm diameter shrinkage in the thrombosed sac (Figs. 3C and 3D). Additionally, neither adverse events nor recanalization of the pseudoAN had been observed in the follow-up period.

Discussion

The course of our case suggests two important clinical issues. First, it serves to remind us that the stump of a cervical ICA occlusion due to chronic post-irradiation injury can progress to a pseudoAN over time, though this is a rare event. In fact, there have been no previous reports of post-irradiated pseudoAN at the carotid stump. The mixed, low echogenic mass observed on carotid US four years previously might have suggested the growth of the thrombosed aneurysm sac at the carotid bifurcation. We speculate that prolonged hemodynamic stress might have accelerated the necrosis of the vessel wall over a long period of time, resulting in an aneurysmal change under the subsequent damage to the vascular endothelial cells by the RT.^{8,9)}

Second, the minimally invasive endovascular approach may have contributed to the thrombosed aneurysm sac shrinkage and mid-term maintenance of the remission at 16 months. Coiling for huge thrombosed aneurysms presents several problems such as incomplete packing, coil compaction, and recanalization.¹⁰⁾ To avoid these, we used mainly hydrogel-coated coils, which offer delayed coil expansion and biocompatibility as inherent technical characteristics.^{11,12)} In addition, we deployed a conventional closed-cell design stent for carotid artery disease across the aneurysmal neck in order to reduce the blood flow to the aneurysm and maintain the vascular patency of the ECA, which was at long-term risk for chronic occlusion.

Recently, endovascular intervention is worth considering as not only an urgent or palliative procedure but also an effective treatment indicated for mid-term remission in the cases of post-irradiated cervical ICA pseudoAN (follow-up period: 3–18 months).^{5–7,13,14)} Additionally, some authors have reported good long-term results of endovascular management for patients with soft-tissue lesions and tumor regression (average survival time: 34 months),¹⁵⁾ as we also observed in our case. Furthermore, it is possible that a covered or flow-diverter stent may be effective against carotid blowout syndrome in an emergent hemostatic situation.^{14,16)}

Nevertheless, there are still many unclear points regarding the treatment strategy for this pathology. The follow-up periods in the available studies have varied widely in length because of the difficulty of managing disease progression in the advanced stages.¹⁷⁾ Therefore, we cannot clarify the radicality of the endovascular approach. Endovascular devices, being foreign matter, can sometimes induce inflammation or immunological reaction when they are implanted in vessels affected by chronic irradiated injury. Furthermore, several major technical complications have recently been reported, including re-bleeding, exposure of the devices due to ulcerated skin associated with infection or tumor necrosis, and acute embolism or delayed cerebral ischemia associated with in-stent thrombosis.^{15,18,19)} As an alternative, surgical resection of the aneurysm and graft bypass operation is often chosen as a radical course of treatment, though this approach also has potential complications including infection and anastomotic leak caused by weakness in the vessel walls and thinning of the skin.²⁰⁾

Since secondary occurrence of pseudoAN can occur in patients whose carotid bifurcations were once totally occluded due to RT, follow-up observation of such patients is required. We suggest performing endovascular intervention in patients with post-irradiated pseudoAN as a minimally-invasive procedure for this advanced clinical disease.

Conflicts of Interest Disclosure

The authors declare that they have no conflict of interest. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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