Lingual Artery-Retromandibular Vein Fistula Four Years after an Uncomplicated Carotid Endarterectomy: Case Report and Review of Possible Etiologies and Treatment Options

Sunil Manjila, MD, Kunal Kumar, MD, Ashish Kulhari, MD, Gagandeep Singh, MBBS, Richard S. Jung, MD, Robert W. Tarr, MD, and Nicholas C. Bambakidis, MD

1Department of Neurosurgery, University Hospitals Case Medical Center, Cleveland, OH, USA
2Department of Neurology, University Hospitals Case Medical Center, Cleveland, OH, USA
3Division of Interventional Neuroradiology, University Hospitals Case Medical Center, Cleveland, OH, USA

Abstract

The external carotid artery’s lingual branch to retromandibular venous fistula following a carotid endarterectomy has not been reported earlier in literature. We report a unique case of an 87-year-old man who had a right-sided carotid endarterectomy in 2009 and presented four years later with complaints of fullness and discomfort in the area of right parotid gland with associated pulsatile tinnitus. A computed tomography (CT) scan of the neck revealed a deep portion of the right parotid gland having abnormal aneurysmal dilatation of a vascular structure, which appeared to be an arteriovenous fistula between branches of right external carotid artery and the retromandibular vein. Conventional catheter angiogram showed a complex arteriovenous fistula seen with the right retromandibular vein receiving multiple small arterial feeders from the right external carotid artery via its lingual artery branch. Slight reflux was noted into the right pterygoid plexus, right maxillary, and right submental veins as well. Surgical treatment was deferred due to high risk of inadvertent facial nerve injury from extensive parotid dissection involved in the procedure. Transarterial embolization of five discrete arterial branches from the right external carotid artery supplying the fistula was performed using particles with resultant remarkable slowing of the venous drainage into the retromandibular vein. After the procedure, his tinnitus and ear fullness resolved completely. The presence of arteriovenous fistula after carotid endarterectomy is a rare yet serious complication and therefore should be diagnosed early and treated promptly. The article highlights the relevant literature on arteriovenous fistula formation in the setting of arterial patch, intraoperative shunting, and surgical-site infections.

Keywords
Arteriovenous fistula; carotid endarterectomy; complications; external carotid-retromandibular vein fistula; pulsatile tinnitus

BACKGROUND

Carotid endarterectomy (CEA) has become a very common and safe procedure for cervical atherosclerotic disease in the past few decades. CEAs are performed routinely in these patients, and delayed vascular complications from CEA are very rare. Pseudoaneurysm is a well-recognized complication after CEA with incidence of less than 1%, although pseudoaneurysms tend to occur in the presence of surgical site infections [1]. Treatment of these challenging pseudoaneurysms has been successfully performed with covered stents such as Wallgraft, Viabahn, and Fluency Plus in post-CEA conditions [2]. Surgical treatment, albeit technically challenging, includes primary angioplasty, partial excision with patch angioplasty, and total excision with interposition grafting, where the basic principles include treatment of infection, vessel reconstruction, and flap coverage if necessary [3]. Interestingly, Yasuda et al., reported a case of infected pseudoaneurysm after CEA with recurrent bleeding, treated with combined surgical and endovascular approach, indicated by severe adhesions of the carotid sheath area. Treatment composed of an initial common carotid to middle cerebral artery bypass using...
radial artery graft, followed by the ligation of distal common carotid artery (CCA), and a subsequent coil embolization of the external carotid artery (ECA) through the facial artery [4]. However, reports of an arteriovenous (AV) fistula after CEA are rarely encountered, with only a handful of cases in literature.

There is a paucity of literature on the etiologies of AV fistula formation in CEA cases as well evidenced by the fact that there is only a single case of delayed carotid-jugular fistula, treated transvenously [5]. The case reports a direct AV fistula, where there is communication between large vessels of the neck, that are located in anatomical contiguity in the carotid sheath. In our case, the patient had undergone an uncomplicated CEA, with an intraoperative shunt and a bovine patch. Patient had no evidence of postoperative infection or neck trauma that predisposed disruption of the vessel wall or development of aberrant vascular collaterals, leading to tinnitus and parotid region swelling. The article examines the role of the above surgical techniques in formation of fistulae, pseudoaneurysms, or arterial dissections, especially while our case had abnormal fistuloust communication between an ECA branch and a venous tributary in the vicinity.

ILLUSTRATIVE CASE

This 87-year-old male patient presented with right-sided pulsatile tinnitus and fullness around the parotid region for several months. The patient had undergone a carotid endarterectomy on the right side four years ago at an outside institution. He was originally diagnosed with 80% right internal carotid artery (ICA) stenosis and subsequently underwent a preoperative angiogram that showed high-grade stenosis 85–90% over 1.5 cm of proximal ICA immediately after carotid bifurcation. A left CCA had irregular atheromatous disease, but no significant stenosis was visualized on arteriography. Intracranial filling of arteries was normal with a hypoplastic right A1 segment. He had medical comorbidities including type-II diabetes mellitus, coronary artery disease with coronary stents, systemic hypertension, and dyslipidemia, apart from prostate cancer and bilateral knee replacements for arthritis. He also had a remote history of melanoma treatment in 1997, details of which were unknown.

Operation

The patient had undergone a right carotid endarterectomy with bovine patch closure at an outside hospital in 2009. Per the operation notes, general endotracheal anesthesia was given, and the patient was positioned supine in a slight beach-chair position with head turned to the left side. About 7500 units of intravenous heparin were administered, and dextran was infused at 100 ml/hr. A standard incision was deepened down to the platysma, medial to the sternocleidomastoid muscle. The right common carotid artery and bifurcation into external and internal carotid arteries were identified. The carotid bulb was low in position; however, the disease was high in the internal carotid artery. The occipital artery branch of the ECA was ligated and dissect to dissection along the course of hypoglossal nerve. Veins in the region were clipped and transected to obtain good exposure of the diseased distal ICA. Vessel loops were used to occlude the arteries in the standard fashion. An arteriotomy was performed and carried up to the bulb, and a Javid shunt was placed in the ICA and secured in the CCA with Javid clamps. An endarterectomy was performed in a smooth subadventitious plane; a tapered end point was achieved in the ICA. After opening the ECA, the bovine patch was then secured to the arteriotomy with 6-0 polypropylene sutures, after removal of Javid shunt and irrigating the lumen with dextran solution. ICA backbleed was obtained to displace air, and the ECA was let open for five cardiac pulsations prior to allowing continuous distal flow through the repaired ICA. After 25 mg of protamine for heparin reversal, a Penrose drain was placed in the depth of the wound and the wound was infiltrated with Marcaine. The wound was closed in layers without complications. No postoperative neck complications were reported.

The patient was followed with serial cerebrovascular Duplex scans performed annually, with no evidence of restenosis with serial demonstration of a widely patent right carotid artery. However, from the beginning of 2013, the patient started having symptoms of right-side ear fullness, pulsatile tinnitus, and right peri-auricular area discomfort. He also had mild neck pain at the surgical site. Considering his remote history of melanoma, he was initially evaluated at an outside institution for a metastatic neck mass. CT of the neck with contrast was done to rule out underlying neoplastic lesion in the region. However, it showed postsurgical changes in the right CCA bifurcation and proximal right ICA, both remaining widely patent. Arterial phase contrast enhancement was noted in the right retromandibular vein, which demonstrated 8 mm × 7 mm aneurysmal dilatation of a vascular structure in the deep parotid gland suggestive of an AV fistula between branches of the right ECA and the retromandibular vein (Fig. 1).

The superficial veins of the left side of the neck did not show similar contrast enhancement or aneurysmal dila-
tation. Parapharyngeal spaces were intact, and parotid–
submandibular glands appeared symmetric and intact;
there was no evidence of cervical lymphadenopathy. The
patient was then transferred to our facility for further
management.

A cerebral angiogram showed a complex A V fistula seen
within the right retromandibular vein receiving multiple
small arterial feeders from the right external carotid
artery and the right lingual artery (Fig. 2).

Slight reflux is noted into the right pterygoid plexus,
right maxillary, and right submental veins. Surgical
treatment was deferred due to high risk of inadvertent
facial nerve injury from extensive parotid dissection
involved in the procedure. Hence, a transarterial emboli-
zation of five arterial fistula branches from the right
external carotid artery was performed using particles
with resultant slowing of the venous drainage into the
retromandibular vein (Fig. 3).

There was residual slightly early venous drainage of the
AV into the right retromandibular vein via a very small
arterial feeding branch from the right lingual artery (Fig.
4).

After the procedure, the patient reported resolution of
pulsatile tinnitus and right ear fullness. He was subse-
quently discharged one day after the procedure.

**Postprocedural course**
Postprocedural follow-up after two weeks reported an
absence of tinnitus and resolution of right ear fullness.

**DISCUSSION**
Carotid endarterectomy has remained a safe and effec-
tive method of treating atherosclerotic carotid artery
stenosis, since the procedure was initially reported by
Eastcott et al in 1954 [6]. Over 100,000 CEA cases are
performed each year in the United States [7–9]; how-
ever, delayed vascular complications are extremely infrequent, especially without trauma or infection. An extensive literature review on delayed vascular complications of CEA revealed only one case of carotid-jugular AV fistula, which was treated transvenously by coil embolization [5]. There have been only a few anecdotes of skull base AV fistula formation reported after carotid endarterectomy; however, most of them were that of carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago. It is interesting to note that the postprocedural AV fistulas reported in early decades were mainly carotid–cavernous fistula reported decades ago.

Iatrogenic AV fistula of the neck occurring between the ECA and EJV is a recognized clinical entity but extremely rare in the setting of a CEA. Rare cases of congenital origin of external carotid-jugular fistulae have also been reported in the literature. Congenital AV fistulas usually appear early in life and are unrelated to trauma [11]. Fistulae involving major vessels such as ECA-JJV, ECA-EJV, ICA-EJV, and ICA-IJV have been reported as rare case reports [12]. However, trauma-related AV fistulas are usually different in presentation. Most patients may remain asymptomatic over a long period, as the presentation is usually insidious, developing over several months to years [13,14]. The most common symptoms related to trauma-related AV fistulas are headache, pulsatile neck mass, and murmur or thrill over...
neck. Fullness in the area of the parotid gland with pulsatile tinnitus is another presentation, as occurred in our patient [5].

Untreated AV fistula the neck could be a potentially fatal condition; if untreated, it may lead to a number of complications with potential long-term effects, including infection, systemic embolization, hemorrhages, and congestive heart failure [13,15]. Therefore, symptomatic AV fistulas should be treated as soon as the patient is diagnosed. Location of these lesions in the neck involving soft tissues renders them to both endovascular and surgical treatment options. Endovascular treatment has received increasing credit in the past years because of minimal invasion and less morbidity and mortality associated with it compared to surgical techniques. Endovascular techniques for obliteration include detachable balloon [16], coil embolization [17], onyx embolization [18], and covered stents like Wallgraft, Viabahn, and Fluency Plus [2,15]. The surgical management in a complex fistula, such as in our patient, would be extremely challenging and would require a partial parotidectomy, with a risk of antecedent facial nerve injury [11].

A review of CEA literature has showed that a patch angioplasty is better than primary closure in CEA procedures with benefits shown in lowering the risk of stroke or death [19], and recurrent restenosis during the perioperative period and on long-term follow-up. Akihito et al proposed that the patch minimizes the effect of neointimal hyperplasia and scarring, maintaining the diameter of treated arterial lumen. Patches have been used in a CEA endarterectomy as early as 1965 [20], while both venous (saphenous vein, with few reported cases of infections) and arterial (locally available superior thyroid artery, or even an occluded femoral artery) wall grafts as well as prosthetic patching materials like polytetrafluoroethylene or Dacron are currently used without additional morbidity of surgical infections. The Dacron patch is a widely used synthetic patch in CEA proce-

Figure 3. Right lingual artery. Selective microcatheter injection revealing the angioarchitecture of the arteriovenous fistula in multiple arterial feeders.
dures, compared to many others. The benefits of a Dacron patch are its availability, resistance to patch-tear and aneurysmal formation, apart from preservation of vein conduits for future potential coronary artery bypass grafting. A Dacron patch is also shown to be at increased risk of infection after CEA procedures. Litwinski et al reported that there is no difference in incidence whether or not the patient undergoing CEA had closure of arteriotomy with a patch angioplasty [3]. Many randomized studies have shown efficacy of patch angioplasty in CEA [21]. The indications include small (<4 mm) ICA; complex, extended, or irregular arteriotomy; and a concurrent distal ICA repair. We use a bovine biomaterial patch, which is also a popular option with its off-shelf availability, biocompatibility, and durability [22,23]. Even when used in infected fields, the bovine patch provides an additional ability to use ultrasound across the patch immediately after grafting. Of note, a less-frequent “eversion endarterectomy” is considered an alternate surgical technique that allows plaque removal without problems of a longitudinal arteriotomy and avoiding patch placement [9].

Besides infection, other important causes of pseudoaneurysm formation after CEA are suture failure, degeneration of the arterial wall, and degeneration of the patch. The pseudoaneurysm due to infection is rare because of very low incidence of post-CEA infection, which is nearly 0.025% to 0.625% [4,24,25]. Staphylococcal organisms are the most common cause of these infections [4,25]. Various factors can cause infection after CEA procedures including long surgery time, repeated or concomitant surgery, incomplete hemostasis, postoperative hematoma, and use of synthetic patches [4,26]. In our case, there was no evidence of any surgical site infections or systemic infections. Pseudoaneurysm due to infection after CEA procedures may present as a localized pulsatile mass, draining sinus, abscess, or hemorrhage and may take two days to eight years after CEA procedures to present [4,24,25,27]. Post-CEA pseudoaneurysms can be managed by surgical or endovascular methods. Surgical management of pseudoaneurysm includes carotid artery ligation, resection of the pseudoaneurysm itself, and patch angioplasty. However, these approaches may be complicated by postoperative stroke, scar formation, and reinfection. Recent developments in the endovascular surgery provide efficacious and safe treatment of pseudoaneurysm. Endovascular methods are associated with less postprocedure complications and allow early recovery after procedures. Various methods can be tried for endovascular treatment of pseudoaneurysm or dissecting aneurysms in the cervical region including stent-assisted coil embolization [4,28], embolization, or detachable balloons [4,18,29]. However, complications associated with the endovascular treatment of pseudoaneurysm of cervical ICA include thromboembolism during or after endovascular procedures, increased risk of persistent infection with coil embolization, and incomplete embolization in patients with irregular-shaped aneurysms.

We feel that the etiology of a delayed AV fistula between branches of ECA and retromandibular vein is...
resultant of cut ends of arteries and veins growing together into a healing scar, and forming de novo connections between the same. Contrary to high-flow ECA-IJV, ICA-IJV, or CCA-IJV iatrogenic fistulae in relation to carotid sheath injury, these lesions are low-flow and delayed in occurrence. On several occasions dictated by anatomical factors, surgeons would sacrifice branches of ECA and local veins in order to access full length of the CCA and its branches, and these extremely rare low-pressure delayed AV fistulae post-CEA would close off spontaneously, never to be clinically recognized or become symptomatic. From the existing body of vascular literature, no direct causation was directed to the use of a shunt or patch during CEA.

**CONCLUSION**

This article portrays the recognition of delayed AV fistulae and endovascular management of this delayed complication after CEA, which presents with pulsatile tinnitus. The authors present this rare scenario to alert the physicians to consider this clinical condition in the differential diagnosis of pulsatile tinnitus with or without parotid swelling after carotid surgery. Regular postoperative follow up of patients with CEA can aid early diagnosis of delayed vascular complications of the procedure like AV fistulae.

**REFERENCES**

26. Durham JR, Malone JM, Bernhard VM. The impact of multiple

