Late-onset Congestive Heart Failure with Multiple Carotid-jugular Fistulae and Pseudoaneurysm After Penetration Injury

Ju-Yi Chen, Shih-Hung Chan, Li-Jen Lin, Chwan-Yau Luo

Trauma-acquired multiple arteriovenous fistulae with late-onset congestive heart failure has not been documented. We describe a 29-year-old man who presented with progressive congestive heart failure 25 years after a penetrating trauma to the left side of the neck. The neck duplex showed a large shunt between the carotid artery and internal jugular vein. Arteriography showed three high-flow arteriovenous fistulae between these two vessels. Magnetic resonance angiography (MRA) showed a carotid pseudoaneurysm originating from three different loci of the carotid artery with a large aneurysm-venous communication between the pseudoaneurysm and the internal jugular vein. Reconstruction of contrast MRA showed three different arteriovenous fistulae, leading to the decision to perform aneurysmectomy, carotid artery repair and jugular vein patch angioplasty. The favorable outcome of this case illustrates that surgery is a reasonable alternative when an endovascular approach is not feasible in patients with trauma-acquired arteriovenous fistulae. [J Formos Med Assoc 2006;105(10):844–847]

Key Words: carotid artery, fistulae, internal jugular vein, pseudoaneurysm

Arteriovenous fistulae involving the major vessels of the neck are uncommon. The majority are traumatic in origin, either due to penetration or to blunt injury. An untreated arteriovenous fistula may lead to a number of complications, the most important of which are heart failure, rupture or emboli. Here, we report the successful treatment of a patient with congestive heart failure caused by high-flow multiple fistulae and a pseudoaneurysm between the left common carotid artery and the internal jugular vein. Broken glass that had penetrated the patient’s neck in a traffic accident 25 years ago had led to the development of this late-onset condition.

Case Report

A 29-year-old man presented with gradual dizziness and dyspnea on exertion. Twenty-five years previously, he had been in a traffic accident in which he suffered a penetrating wound on the left side of the neck that had been treated with primary suturing only. He had, however, shown no significant symptoms or signs since the accident. On this presentation, he appeared healthy, but physical examination found a pulsatile mass on the left side of the neck with a thrill and a continuous bruit, reaching its maximum below the left ear.
Chest X-ray showed cardiomegaly and a clear lung field. Echocardiography showed left ventricular dilatation with a left ventricle end-diastolic diameter of 5.69 cm and a left ventricular ejection fraction of 46%. Color Doppler sonography showed a continuous mosaic flow from the left internal carotid artery to the internal jugular vein. Angiogram of the left carotid artery demonstrated a huge communication between the carotid artery and the jugular vein, in which there was simultaneous opacification of a dilated left internal jugular vein, so that it did not precisely delineate the locations of the communications. Magnetic resonance angiography (MRA) showed that the carotid artery was twice its normal size, that the internal jugular vein was nearly four times its normal size, and that there was a mass lesion in between (Figure 1A). Further reconstruction of the MRA image revealed this mass to be a pseudoaneurysm with multiple drainage from the carotid artery and a large base into the internal jugular vein (Figure 1B). There were three fistula tracks from the carotid artery into the pseudoaneurysm: the first drained from the distal part of the common carotid artery, the second drained from the bulb of the carotid artery and the third drained from the left internal carotid artery (Figure 2).

Due to symptoms of congestive heart failure, intervention was suggested. After considering the complexity of these multiple fistulae and the pseudoaneurysm, the possible complications of endovascular treatment, and the surgical feasibility of the anatomic location and characteristics, surgical division of the arteriovenous fistulae tracts and excision of the pseudoaneurysm were recommended. Under general anesthesia, the left side of his neck was explored via an incision along the anterior border of the sternocleidomastoid muscle. Under the sternocleidomastoid muscle, which was laterally displaced, the internal jugular vein

![Figure 1](image1.png)  
**Figure 1.** (A) Magnetic resonance angiography (MRA) shows a large carotid artery (white arrowhead), huge internal jugular vein (white arrow) and an unidentified mass-like lesion (black arrow) between these two vessels in the posteroanterior view. (B) Reconstructed MRA shows a pseudoaneurysm (white arrow) with multiple fistulae tracts (black arrows) connecting the carotid artery and a broad base (19 mm in diameter) (dotted line) draining into the internal jugular vein in the left lateral view.

![Figure 2](image2.png)  
**Figure 2.** Reconstructed magnetic resonance angiography shows three fistulae tracts (black arrows) connecting the carotid artery and pseudoaneurysm (white arrow): (A) right oblique view; (B) left oblique view.
(diameter, 15 mm) and its branches were distended. The aneurysm (4 x 3.5 cm) was on the anterolateral side of the internal jugular vein. A firm fibrous mass composed of fistular tracts was also found between the carotid artery and pseudoaneurysm. The internal jugular vein and the common, external and internal carotid arteries were explored and controlled proximally and distally. Because the intraoperative carotid pressure distal to the clamp was > 50 mmHg, an internal shunt was not used for carotid artery repair. The fibrotic mass, which was composed of the fistula tracts, and the pseudoaneurysm between the carotid artery and internal jugular vein were resected. Following debridement of the surrounding fibrotic tissue, the orifices of the fistula tracts to the aneurysm laterally over the carotid artery were repaired directly. Then, because half the circumference of the jugular vein was resected with the aneurysm, the internal jugular vein was reconstructed with an autogenous venous graft patch. The neck thrill disappeared immediately after surgery.

The patient recovered uneventfully and was discharged 6 days after surgery. He suffered no adverse sequelae from the surgical division, and remained asymptomatic with complete resolution of the dyspnea. Three months later, Doppler ultrasound of the carotid artery showed no evidence of fistula recurrence, and echocardiography showed that the left ventricle end-diastolic diameter had decreased to 5.2 cm and the left ventricular ejection fraction was 55%. He was healthy at the 12-month follow-up.

Discussion

Traumatic arteriovenous fistulae of the head and neck account for less than 4% of the complications consequent to arterial trauma in these areas. The majority of cases are due to complications resulting from gunshot wounds, stabbing or iatrogenic injuries for therapeutic purposes. Occasionally, this condition may result from blunt neck trauma with or without bony fracture. Penetrating wounds involving the carotid arteries can lead to two possible and important late sequelae: pseudoaneurysm and arteriovenous fistula. In this case, an artery and adjacent jugular vein were simultaneously lacerated by broken glass in a traffic accident. This led to the formation of a pseudoaneurysm after the arterial wall sustained multiple punctures and blood extravasated into a compartment. The pseudoaneurysm communicated with both artery and vein via three fistulae from the carotid artery to the internal jugular vein. If untreated, arteriovenous fistulae or carotid artery pseudoaneurysm may cause congestive heart failure, cerebral ischemia, thromboembolism or even rupture complications. A single opening is always the rule for fistulae of traumatic origin, whereas congenital fistulae usually have multiple arteriovenous communications. Multiple fistulae of traumatic origin with drainage into a pseudoaneurysm and with high-cardiac-output heart failure caused by large left-to-right shunt as occurred in this case have not been previously documented in the English literature.

Appropriate detection and evaluation of arteriovenous fistulae requires, in addition to detailed history taking and physical examinations, the use of diagnostic tools such as noninvasive color Doppler sonography, contrast computed tomography angiography or contrast MRA, or invasive angiography. Color Doppler sonography is a non-invasive diagnostic means of screening patients suspected of having an arteriovenous fistula, and is a convenient follow-up method after intervention. However, more detailed information about the structures, extension, complexities and relationships of the fistulae with local tissues are essential when considering the most appropriate therapeutic intervention. Although angiography remains the standard of care because of its reported 100% accuracy, it may fail to delineate the exact loci of communication when a huge shunt develops, as in the present case. Reconstruction of contrast MRA is a noninvasive, convenient and timesaving technique that can provide detailed information about the structures of the fistulae and pseudoaneurysms. This technique not only influenced the decision on the mode of treatment in this case, but also aided preoperative planning.
Treatment options for carotid arteriovenous fistulae include operative repair, detachable balloon, coiling or stenting. In this case, there were some concerns about endovascular treatment despite recent technological advances in endovascular methods and devices, which may reduce the occurrence of these problems. Although successful treatment of carotid-jugular fistula using a detachable balloon has been reported, some clinicians consider this suitable only for fistulae at the cranial base and as an adjunct to surgery. In addition, the frequency of recurrent fistulae (13%) caused by premature balloon deflation and migration or bony-fragment puncture of balloons after embolization is relatively high. Second, deploying the balloon to the target lesion site in patients with three different orifices resulting in a high-flow shunt, as in this case, may be difficult. Third, multiple-coil embolization with high-flow fistulae may risk dislodgement of the coils, which would lead to a pulmonary embolism or stroke. Fourth, a patient undergoing such treatment has to tolerate multiple instances of transient cerebral ischemia for carotid artery temperate occlusion in order to deploy multiple coils, increasing the risk of undesired sequelae of brain ischemia.

Covered stent grafts have recently been successfully used to treat traumatic carotid fistulae and pseudoaneurysms. Previously, however, this use had been limited due to concerns about the risk of a cerebral infarction resulting from acute dissection, occlusion or embolic sequelae of stent grafts. The presumed risk of latent occlusion from thrombus formation or intimal hyperplasia is an additional concern. Smith et al reported delayed stenosis (12–18 months) following placement of a covered wall graft endoprosthesis in three cases of carotid artery pseudoaneurysm. Thus, the use of carotid stent grafts raises questions about long-term patency and freedom from restenosis, especially in a young patient. In the present case, the complexity of the multiple fistulae revealed by the reconstructed MRA images suggested that nonsurgical therapeutic techniques would be more difficult, have a low probability of success and increase the risk of embolic stroke. This led to the decision that surgical intervention was the best therapeutic choice.

In conclusion, this case illustrates that noninvasive contrast MRA should be arranged for patients with fistulae of traumatic origin when a traditional angiogram demonstrates high-output arteriovenous communication. The use of this technique is necessary to determine the detailed structures of the fistulae to enable precise preinterventional evaluation for either surgery or endovascular techniques. Due to the risk of cerebral ischemia when using a detachable balloon or coil, concerns about restenosis when using an endovascular covered stent graft, and the difficulty of anatomic correction, open repair may be the best choice for patients with complex fistulae of traumatic origin.

References