The Potentiality for Development of Multiple Dural Arteriovenous Fistulas after Ligation of the Internal Jugular Vein: A Case Report

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A 74-year-old male presented with an intracranial hemorrhage caused by multiple dural arteriovenous fistulas (DAVFs) in the left transverse sinus and right sigmoid sinus. Four months previously, the patient underwent tongue cancer removal with lymph node dissection and ligation of the right internal jugular vein. Endovascular embolization (transvenous and transarterial embolization) resulted in the complete disappearance of the fistulas. Follow-up angiography revealed new arteriovenous shunts at the superior sagittal sinus and right transverse sinus, and we treated the patient with staged transarterial embolization. Finally, venous congestion almost completely resolved and the DAVFs disappeared without any sign of recurrence. This case speculates the concept of DAVF as an acquired lesion caused by intravenous hypertension and alerts clinicians to take precautions against ligation of the internal jugular vein during a cervical operation.

**Keywords:** dural arteriovenous fistula, venous hypertension, internal jugular vein, endovascular surgery

**Introduction**

The etiology of dural arteriovenous fistula (DAVF) remains unclear. Generally there are two major categories, congenital and acquired. Recent reports have suggested that DAVFs are acquired anomalies caused by venous hypertension.¹ ¹² We herein report a rare case of multiple DAVFs that have the potential to develop after ligation of the internal jugular vein during a cervical operation.

**Case Report**

A 74-year-old male became unconscious with the meningeaL irritation sign on September 14, 2011. On September 19, the patient’s consciousness worsened and computed tomography (CT) scanning showed an intracranial hemorrhage in the left cerebellar hemisphere and a small acute hematoma in the left frontal epidural space (Figs. 1A and 1B). Cranial magnetic resonance imaging (MRI) and enhanced CT scanning revealed cortical venous dilatation with significant brain edema on the posterior cranial fossa (Figs. 1C and 1D). Cerebral angiography revealed multiple DAVFs in the left transverse sinus and right sigmoid sinus (Cognard type IIa+b) (Fig. 2).

The patient’s past history was remarkable for tongue cancer removal with lymph node dissection and ligation of the right internal jugular vein 4 months previously. Preoperative enhanced CT scanning revealed that his right jugular vein was twice the size of the left jugular vein (Fig. 3A) and there were no venous dilatations on the posterior lobe (Fig. 3B). The right jugular vein was not enhanced in postoperative CT scanning.

On September 21, we performed endovascular embolization. First, transvenous embolization was performed to the venous pouch with an arteriovenous shunt at the right sigmoid sinus. Although we attempted to embolize the parasinus arteriovenous shunt at the left transverse sinus via transvenous and transarterial embolization through the left occipital artery, the arteriovenous shunt was not reduced. Eventually, coil embolization from the near confluence to just before the vein of Labbe was performed, for the arteriovenous shunt at the left transverse sinus. Finally, we achieved complete disappearance of the fistulas with improved venous congestion. Gradually, the patient’s consciousness improved.

In March 2012, follow-up angiography revealed newly acquired multiple DAVFs at the superior sagittal sinus and right transverse sinus. Although staged transarterial embolization was performed, recurrence at the superior sagittal sinus and new shunts at the left sigmoid sinus and left cortical vein were revealed. Transarterial embolization was performed, and the retrograde venous flow and dilatation of the cortical vein disappeared. Follow-up angiography showed that the fistula at the transverse sinus disappeared spontaneously, and the anterograde arteriovenous shunts at the left sigmoid sinus and left cortical vein remained (Fig. 4).

**Discussion**

Recent reports have generally suggested that most DAVFs are caused by acquired lesions, venous hypertension, sinus thrombosis, trauma, intracranial infection, or the occlusion of the dural sinuses. Previous research suggests that venous hypertension can be more precisely and closely related to the pathogenesis of acquired DAVF than the other factors, consistent with the results of an experimental rat model and clinical observations.¹ ¹²

Resection of the internal jugular vein can be necessary in cases of radical neck dissection and is not challenging in many cases with the development of collateral circulation. Previous
literature reported that the loss of the internal jugular vein is associated with morbidity, including increased cerebral edema, stroke, laryngeal edema, blindness, facial fullness, and dural thrombosis.\textsuperscript{3,4} There were only two cases which suggested that venous hypertension caused by stenosis or the occlusion of the internal jugular vein induced DAVFs.\textsuperscript{3,4} The presented case was potentially “de novo” DAVFs because there were no venous dilatations before the ligation of the internal jugular vein and newly acquired DAVFs were revealed after the first interventional surgery. Although, this case has the possibilities that DAVFs with no retrograde flow already formed before the operation of the tongue cancer and became clinically evident after the occlusion of the internal jugular vein and the blockage of the anterograde flow. Ligation of the right jugular

![Fig. 1 CT scanning reveals an intracranial hemorrhage in the left cerebellar hemisphere (A) and a small acute hematoma in the left frontal epidural space (B). Cranial MRI shows significant brain edema and high-intensity signals around the cerebellar sulcus on FLAIR images (C). Enhanced CT scanning reveals abnormal venous dilatation on the posterior lobe (white arrow) (D).](image1)

![Fig. 2 Anteroposterior view (A) and lateral view (B) of the left external carotid angiograms before endovascular surgery demonstrate one arteriovenous shunt (white arrows). The arteriovenous shunt is fed by the left middle meningeal artery, bilateral occipital artery and left posterior meningeal artery and drained into the left transverse sinus. Lateral view of the left internal carotid angiograms in the venous phase (C) shows that the venous flow drains through the left sigmoid sinus into the internal jugular vein with high-grade venous congestion. Anteroposterior view (D) and lateral view (E) of the right external carotid angiograms reveal another arteriovenous shunt with venous pouch (white arrows). The shunt at the right sigmoid sinus is fed by the right occipital artery and right ascending pharyngeal artery with retrograde flow into the superior sagittal sinus. Lateral view of the right internal carotid angiograms in the venous phase (F) shows that the venous flow drains through the right cavernous sinus and pterygoid plexus with high-grade venous congestion.](image2)

![Fig. 3 Enhanced CT in the venous phase of pre-ligation of the right internal jugular (A) vein demonstrates that the right jugular vein (white arrow), greatest dimension of 24.3 mm, was twice the size of the left jugular vein (black arrow). There were no venous dilatations on the left posterior lobe (B).](image3)
vein, which is clearly the dominant side and main drainage of the intracranial blood flow, may act as a trigger, changing the local hemodynamics and producing venous hypertension. Our case formed multiple DAVFs earlier than the previous case. Modifying the venous drainage on the dominant side of the internal jugular vein induced the opening of the “dormant” channels and led to exacerbation of the arteriovenous shunts. In addition, venous hypertension also induced chronic regional hypoperfusion and the expression of angiogenic factors, which stimulated vascular endothelial proliferation and excessive angiogenesis in the dura mater around the sinus.

The present case showed that newly acquired multiple DAVFs emerged at a different site after first embolization. Our findings suggested that correction of the venous flow and normalization of venous hypertension were important to plan the surgical procedure, as well as ligation of the shunt points. Fukumoto et al. reported a DAVF case with sinus stenosis that was completely treated by a stenting procedure. If venous hypertension was caused by sinus stenosis, stenting technique was in the options. It is difficult to treat with stenting in the acute stage such as the presented case with cerebral hemorrhage. Although rat models are anatomically distinct from humans, our case established the possibilities of a connection between venous hypertension and the formation of DAVF in humans and exemplified the importance of acquired factors.

**Conclusion**

We herein reported a rare case of multiple DAVFs associated with venous hypertension after ligation of the right jugular vein. Our case suggested that an acquired lesion and multiple acquired factors, particularly venous hypertension and activation of angiogenic factors, played important roles in the formation of DAVFs. When performing ligation of the internal jugular vein during a cervical operation, we should keep in mind the possibility that ligation of the internal jugular vein may lead to the formation of a DAVF.

**Conflicts of Interest Disclosure**

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**References**


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