

Endarterectomy of carotid artery bifurcation in the setting of a persistent hypoglossal artery and anomalous collateral vascular supply

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ABSTRACT

Presented is a patient with carotid artery stenosis resulting in crescendo anterior and posterior circulation transient ischemic attacks. Treatment was complicated by a rare persistent hypoglossal artery (HGA) arising from the left internal carotid artery in addition to severe contralateral carotid disease, hypoplastic vertebral arteries, and incomplete circle of Willis. A carotid endarterectomy with shunting was performed, maintaining perfusion of both the proper left internal carotid artery and HGA. This is a rare case of carotid stenosis in the setting of a persistent HGA with contralateral carotid disease and highlights the importance of planning intracranial perfusion before carotid surgery. (*J Vasc Surg Cases and Innovative Techniques* 2020;6:520-3.)

Keywords: Vascular surgery; Hypoglossal artery; Carotid endarterectomy; Shunt

CASE REPORT

A 79-year-old right-handed woman presented with three episodes of aphasia, right arm weakness, and vertigo with dysmetria during 6 days. The episodes occurred spontaneously at rest without association with activity or medications and resolved after several minutes. She had no history of similar events and was asymptomatic on presentation. She was admitted for crescendo transient ischemic attacks involving the left hemisphere and probably the cerebellum.

Her past medical history was significant for type 2 diabetes on oral antihyperglycemics, dyslipidemia managed with a statin, hypertension controlled with hydrochlorothiazide, and smoking. She lived independently. On physical examination, a palpable thrill over the left carotid and weak right carotid pulse were noted. The neurologic examination findings were grossly normal, with normal neck range of motion. She was normotensive and in normal sinus rhythm with no significant cardiac or respiratory findings.

Computed tomography angiography demonstrated an 80% stenosis of the left carotid artery bifurcation. The plaque extended 22 mm into the internal carotid artery (ICA). A persistent left hypoglossal artery (HGA) bifurcated from the left ICA

12 mm distal to the end of this plaque (34 mm from the origin of the ICA). The HGA was of equivalent caliber to the proper left ICA and formed the main supply to the basilar artery (Fig 1). Accordingly, the left vertebral artery terminated in the posterior inferior cerebellar artery, and the right vertebral artery was hypoplastic. The right carotid bifurcation was severely diseased and probably occluded, with the right internal and external carotid arteries reconstituting distally (Fig 2). Intracranially, posterior communicating arteries were absent on the left hypoplastic on the right. Subsequent magnetic resonance imaging did not reveal appreciable cerebral or cerebellar infarcts.

The patient was aware of the relatively higher risks of intervention because of her anomalous anatomy. Endovascular management of the left carotid bifurcation stenosis was avoided from the femoral approach because of significant nonocclusive aortic arch atherosclerosis and from the femoral or transcarotid approach because of the proximity of the carotid lesion to the HGA origin and concern of protecting both the proper ICA and HGA.

Left carotid endarterectomy was performed under general anesthesia with intraoperative shunting the day after her presentation to the hospital. The distal extent of the ICA atherosclerotic lesion terminated at the level of the C2 vertebral body. The hypoglossal nerve was mobilized to facilitate exposure of the diseased ICA. Dissection continued superiorly to the level of the C1 vertebral body to selectively control both the proper ICA and HGA. This was facilitated by dividing the posterior belly of the digastric and retracting the stylohyoid muscle to expose the origins of the proper ICA and HGA (Fig 3).

Adequate normal artery for placement of a ring clamp on the ICA distal to the plaque and proximal to the HGA origin was confirmed, followed by arteriotomy and immediate Argyle shunt placement 2 cm into the proper ICA (Fig 4). Placement of the distal tip of the shunt proximal to the HGA origin was achieved by palpation and intraoperative Doppler interrogation

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Fig 1. Persistent hypoglossal artery (HGA). This right posterior oblique view demonstrates the standard three great vessels arising from the aortic arch (*right to left*: innominate, left carotid, left subclavian). The subclavian arteries have been truncated. Distal to the left carotid bifurcation, the persistent HGA arises from the internal carotid artery (ICA). The HGA takes a tortuous path to become the primary supply to the basilar artery.

to ensure antegrade perfusion to both the HGA and proper ICA. This was critical because of diminutive vertebral or contralateral carotid collaterals coupled with an incomplete circle of Willis. After carotid endarterectomy, a Dacron patch closure was performed with removal of the shunt before completion of the closure (Fig 5). The patient had no neurologic deficits after surgery and no recurrent neurologic events or residual stenosis on duplex ultrasound examination 1 month postoperatively. Consent was obtained by the patient to publish this case.



Fig 2. Concurrent extracranial cerebrovascular disease. The true left carotid bifurcation is significantly stenosed with dense calcification. Distally, the hypoglossal artery (HGA) arises from the left internal carotid artery (ICA) and courses through the foramen magnum to form the basilar artery. There are no large vertebral arteries communicating with the basilar artery. The right common carotid artery is diminutive and probably occluded at the bifurcation.

DISCUSSION

This case highlights the challenges of managing carotid artery bifurcation stenosis in the setting of a persistent HGA. A persistent HGA is a type of persistent fetal carotid-basilar anastomotic artery, which are embryologic remnants that fail to regress in utero by the sixth embryologic week.^{1,2} Persistent carotid-basilar anastomotic arteries in adults are rare and therefore their prevalence is difficult to estimate. In a review of 4400 adult cerebral angiograms, persistent carotid-basilar anastomoses were seen in 0.13% of patients including one (0.023%) persistent HGA.³

Persistent HGAs are often accompanied by hypoplastic or atretic vertebral and posterior communicating arteries.⁴⁻⁶ In the setting of a persistent HGA, at least one vertebral or posterior communicating artery is hypoplastic or atretic in 86% and 94%, respectively, and both are hypoplastic or atretic in 28% and 78%, respectively.⁵ These findings have been noted by previous authors as complicating carotid endarterectomy in the context of a persistent HGA.^{7,8}

Because a persistent HGA often forms the sole supply of the basilar artery and posterior circulation, high flow through the ipsilateral carotid artery bifurcation is known to result in false carotid stenosis diagnoses on

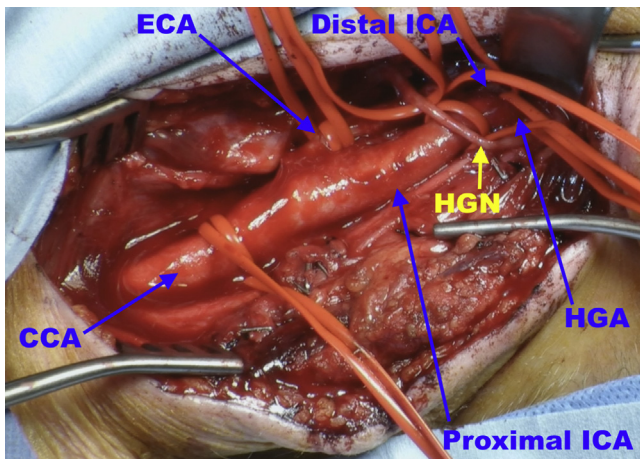


Fig 3. Intraoperative photograph of the left carotid artery dissection; before arterial clamping. Cranial orientation is to the top right of the image. The common carotid artery (CCA), external carotid artery (ECA), and proximal internal carotid artery (ICA) form the typical carotid bifurcation. The hypoglossal nerve (HGN) has been skeletonized. Cranial to the hypoglossal nerve, the individual origins of the hypoglossal artery (HGA) and distal ICA were controlled with Silastic vessel loops.

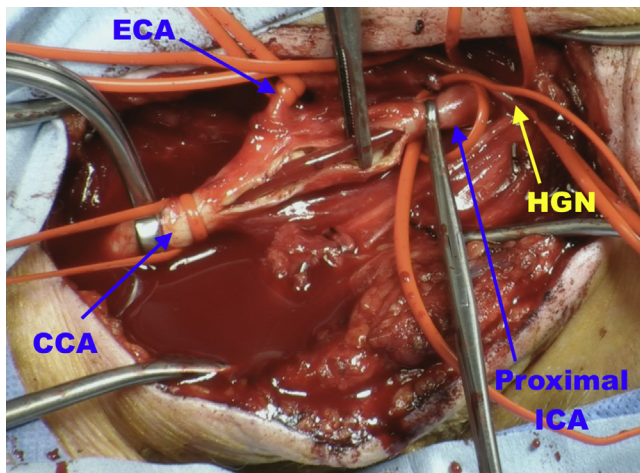


Fig 4. Intraoperative photograph of the left carotid artery dissection; application of shunt. After arteriotomy, an Argyle shunt was placed extending from the common carotid artery (CCA) proximally to the proximal internal carotid artery (ICA) distally. We manually palpated and Doppler interrogated the hypoglossal artery (HGA) and distal ICA to ensure that the distal extent of the shunt was proximal to the origin of the HGA. This was to maintain perfusion to both branches and to not inadvertently occlude the primary vessel perfusing either anterior or posterior circulation. ECA, External carotid artery; HGN, hypoglossal nerve.

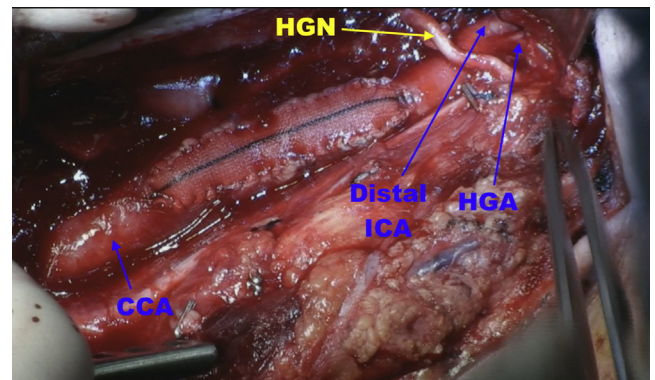


Fig 5. Intraoperative photograph of the left carotid artery dissection; completion after endarterectomy and Dacron patch closure. CCA, Common carotid artery; HGA, hypoglossal artery; HGN, hypoglossal nerve; ICA, internal carotid artery.

The intracranial vascular anomalies that often accompany a persistent HGA pose specific challenges in the surgical management of carotid artery bifurcation disease and require thorough preoperative planning. In this case, essentially all intracranial perfusion was supplied through the left common carotid artery stenosis, corroborated by the strongly palpable left carotid thrill. Furthermore, the anterior and posterior circulations were independent because of diminutive posterior communicating arteries. These features required maintenance of antegrade perfusion of both the proper ICA and HGA during carotid endarterectomy through shunting.

The need for shunting during carotid endarterectomy in the presence of anomalous cerebral and cerebellar collateralization is also complicated by the typical proximity of the origin of the HGA with the origin of the ICA, between C3 and C1.¹ Because this patient also had significant contralateral carotid disease, maintaining perfusion through both the HGA and the proper ICA was essential. In previous reports of carotid endarterectomy in the presence of an HGA requiring shunting, five selectively cannulated the HGA,^{7,10,14-16} one selectively cannulated the proper distal ICA,¹⁷ and six cannulated the proximal ICA caudal to the HGA.^{11,18-22} Cartier et al²⁰ noted that despite shunting, the plaque extended to the origin of the HGA, which required temporary shunt removal to facilitate endarterectomy. Kawabori et al¹¹ noted that the shunt was easily placed too distal and interrupted flow to one of the two branches, requiring intraoperative angiography to confirm their shunt placement. Similarly, the shunt in this case report initially passed into the HGA and needed to be withdrawn into the proximal ICA to maintain anterior and posterior cerebral perfusion.

Alternatively, carotid stenting of the carotid bifurcation and proximal ICA with duplicate distal protection devices in both the proper ICA and HGA has been

the basis of ultrasound velocity criteria.⁹ Our patient presented with both anterior and posterior circulation symptoms, which has been reported by five previous authors.^{4,10-13}

reported.²³ This technique requires jailing of one of the distal protection devices outside the stent, which needs to be withdrawn on completion. The use of flow arrest or reversal protection such as transcarotid artery revascularization in this case would not be ideal because of the poor collateral circulation. Furthermore, the proximity of the HGA origin to the ICA origin and the frequent tortuosity of the HGA may complicate endovascular treatment.¹

CONCLUSIONS

This is a rare case of carotid artery stenosis in the setting of a persistent HGA with severe contralateral carotid artery disease that emphasizes the importance of preoperative planning of intracranial perfusion during carotid surgery.

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