Rare Variant of Persistent Primitive Hypoglossal Artery, Arising From the External Carotid Artery

—Case Report—

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Abstract

A 63-year-old man presented with an extremely rare variant of persistent primitive hypoglossal artery (PHA), which was found incidentally during examination for a contralateral asymptomatic internal carotid artery (ICA) stenosis. This anastomotic vessel arose from the external carotid artery, not the ICA, and joined the vertebrobasilar artery through the hypoglossal canal. Persistent PHA is rare and the reported incidence is 0.027–0.26%. Recognition of the existence of this variant vessel and preservation during neuroradiologic intervention or surgery is important to prevent possible ischemic complications.

Key words: persistent hypoglossal artery, external carotid artery, carotid-vertebrobasilar anastomosis, rare variant, hypoglossal canal

Introduction

Persistent primitive hypoglossal artery (PHA) is a rare embryonic carotid-vertebrobasilar artery anastomosis with a reported incidence of 0.027–0.26%.2) The hypoglossal artery usually arises from the internal carotid artery (ICA) between the C1 and C3 levels and traverses through the hypoglossal canal to join the vertebrobasilar artery. We present an extremely rare case of a variant of the persistent PHA arising from the external carotid artery (ECA).

Case Report

A 63-year-old man was admitted with an asymptomatic left ICA stenosis. Magnetic resonance (MR) angiography, three-dimensional computed tomography (3D-CT) angiography, and conventional angiography to evaluate the cervical and intracranial vessels identified an anomalous vessel arising from the right ECA at the C3-4 level and joining the vertebrobasilar artery (Fig. 1). Time-of-flights imaging, source imaging of 3D-CT angiography, and 3D rotational angiography demonstrated that this anomalous vessel had entered the skull through the hypoglossal canal (Fig. 2). The right vertebral artery was hypoplastic and this anastomotic vessel joined the intradural right vertebral artery (Figs. 3 and 4). Intracranial MR angiography revealed normal connection of the bilateral intradural vertebral arteries and basilar artery (Fig. 4). The patient underwent left carotid endarterectomy (CEA) successfully and was discharged uneventfully. Medical antiplatelet therapy has been continued.

Discussion

The primitive trigeminal, otic (acoustic), hypoglossal, and proatlantal intersegmental arteries are persistent fetal anastomoses between the carotid and vertebrobasilar circulations. These fetal anastomoses emerge at the 4 to 5 mm embryonic stage, persist for approximately 1 week, and regress at roughly the rate at which the posterior communicating and vertebral arteries are developed. The first to disappear, and the rarest, is the otic artery, followed by
Fig. 2 Time-of-flight image (A), source image of the three-dimensional computed tomography angiography (B), and three-dimensional rotational angiogram (C) revealing the anomalous vessel passing through the hypoglossal canal (arrow).

Fig. 3 Cervical magnetic resonance angiograms showing the hypoplastic right vertebral artery (arrow).

Fig. 4 Intracranial magnetic resonance angiograms revealing an anomalous vessel connecting to the intradural right vertebral artery (arrow).

the hypoglossal artery, the trigeminal artery, and then the proatlantal intersegmental artery. The proatlantal intersegmental artery maintains the posterior circulation until the vertebral arteries are fully developed between 7 and 8 weeks. Occasionally, persistence of these vessels is noted after birth and into adult life.

The diagnosis of persistent PHA is based on the following criteria: the artery arises at C1 to C3 as a robust branch of the ICA; follows a somewhat tortuous course, then proceeds through the hypoglossal canal to the posterior cranial fossa, and does not traverse the foramen magnum; the basilar artery is filled only beyond the anastomosis; and the posterior communicating arteries are absent (i.e., not visible on angiography). In addition, the vertebral artery is aplastic on the ipsilateral side and hypoplastic on the contralateral side, or hypoplastic on both sides. The proatlantal artery arises from the ECA, the ICA between the C2 and C4 levels, or the common carotid artery (CCA) bifurcation, and joins the vertebral artery via the foramen magnum. One of the most important factors differentiating these two anastomoses is whether the vessel enters the hypoglossal canal. In the present case, the anastomotic vessels arose from the ECA, passed through the hypoglossal canal, and joined the vertebrobasilar artery. Therefore, this case was considered to be a rare variant of the persistent PHA.

Only four similar cases have been reported previously. A 68-year-old man with left CCA stenosis of 50% just proximal to its bifurcation had 80% stenosis of the proximal ECA at the origin of the persistent PHA, and underwent left CEA. Recognition of this vessel and its preservation are probably important during surgery. A 59-year-old man presented with a right ICA aneurysm and left vertebral artery aneurysm, which were successfully clipped, and a persistent PHA arising from the left ECA. A 60-year-old man presented with left thalamic hemorrhage and an unusual variant of persistent left PHA. Finally, a 78-year-old man had a rare variant of persistent left PHA which was found incidentally during an examination for dizziness.

The embryological anatomy of the hypoglossal artery arising from the ECA involves the ascending pharyngeal artery which normally arises from the proximal ECA and divides into two major trunks (pharyngeal and neuromeningeal trunks). The neuromeningeal trunk branches into the hypoglossal canal. This hypoglossal branch of the ascending pharyngeal artery may be a remnant of the PHA. The failure of this connection to involute was probably the cause of the present unusual variant of the persistent PHA arising from the ECA.

If the PHA persists, the ipsilateral vertebral artery and posterior communicating artery are hypoplastic, and the contralateral vertebral artery and posterior communicating artery are only present in one-third of the cases. Therefore, the persistent PHA is the main or exclusive feeder of the posterior circulation. Temporary clamping of the persistent PHA during CEA may increase the risk of ischemia. Furthermore, the ECA may be ligated for several reasons. Wounds, resection of the upper jaw, hemorrhage from the tonsils, and cutting of the blood supply for hyper vascular tumor or intractable epistaxis may also increase the risk of ischemia.

Some clinical symptoms associated with the persistent PHA include hypoglossal nerve palsy, as well as neuralgia of the glossopharyngeal nerve due to irritation by the large persistent PHA. However, the persistent PHA is mostly found incidentally at angiographic evaluation. Although conventional angiography is considered the standard of reference when evaluating vascular structures, MR and 3D-CT angiography are noninvasive alternatives that have some advantages. In the present case, the variant of the persistent PHA was found on MR, 3D-CT, and conventional angiography incidentally during an evaluation for contralateral ICA stenosis. Recently, these diagnostic tools have been improved and can afford excel-

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lent anatomic localization of the craniocervical vessels. Further improvements in diagnostic technology, including advances in equipment and imaging techniques, may detect more frequent diagnosis of this extremely rare variant of persistent PHA.

Recognition of the existence of this variant vessel and preservation during neuroradiologic intervention or surgery is important to prevent possible ischemic complications.

References


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