Incidentally Detected Persistent Hypoglossal Artery Detected on Magnetic Resonance Imaging and Angiography: A Rare Vascular Anomaly

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Abstract

We report a rare case of persistent primitive hypoglossal artery detected incidentally on MRI and catheter angiography in a patient with falx meningioma with review of literature.

Keywords: Persistent primitive hypoglossal artery; Digital subtraction angiography; Magnetic resonance imaging; Carotid-basilar anastomoses; Internal carotid artery; Basilar artery; Vertebral artery

Commentary

A persistent primitive hypoglossal artery (PPHA) is rare vascular anomaly with an incidence of 0.02%-0.26% [1,2]. PPHA is the second most common carotid-vertebrobasilar artery anastomosis after trigeminal artery [2]. PPHA is usually an incidental finding in most of the cases however it may be associated with intracranial aneurysm, arteriovenous malformation, glossopharyngeal neuralgia and hypoglossal palsy [3-6]. We report a rare case of incidentally detected PPHA in patient with midline falx meningioma diagnosed on magnetic resonance imaging (MRI) as aberrant flow void structure (artery) coursing through the right hypoglossal canal. It was later confirmed on digital subtraction angiography (DSA) of brain and neck vessels.

A 40 year old male patient presented with history of chronic daily headache for 5 months which was not controlled with medications. MRI was done which showed a falx meningioma and an incidental anomalous artery with flow void coursing through the right hypoglossal canal (Figures 1A and 1B). Patient was advised surgical resection of the tumor. In view of atypical imaging findings for meningioma, DSA was planned preprocedural to assess the tumor vascularity and to perform endovascular embolization if required. Right internal carotid artery (ICA) injection showed an anomalous artery arising from the cervical segment of ICA at C2-C3 vertebral level coursing posteriorly and superiorly to enter intracranially and continuing as basilar artery (Figures C and D). Bilateral subclavian artery injection showed hypoplastic bilateral vertebral artery (VA) (Figures 1E and 1F).

Discussion

When the developing human embryo reaches a diameter of approximately 4 mm, the ICAs extend and anastomose at three major sites with the paired longitudinal neural arteries which correspond to primitive vertebrobasilar system in the hindbrain. These arteries include the trigeminal, otic, and hypoglossal arteries and are named according to their neighboring structures. There are seven transversely oriented arteries in the cervical region. The most cephalic of these...
Common of all carotid-vertebrobasilar anastomoses following embryonic development results in various persistent carotid-vertebrobasilar anastomosis including persistent trigeminal artery, primitive hypoglossal artery, proatlantal intersegmental artery, persistent otic artery, persistent dorsal ophthalmic artery, and persistent primitive olfactory artery [7]. The PPHA is second most common of all carotid-vertebrobasilar anastomoses following persistent trigeminal artery and usually arises from the cervical ICA at the level of C1-C2 vertebral bodies but never below C3-C4 level. In our case, it arose from the cervical ICA at the level of C2-C3 level. It then courses through hypoglossal canal along with the accessory nerve and anastomosis with the basilar artery. A PPHA is commonly associated with aplastic or hypoplastic VA, posterior communicating artery, posterior cerebral artery and/or anterior inferior cerebellar artery. Thus, the findings of aplasia or hypoplasia of these vessels should also raise the suspicious of carotid basilar anastomosis [8-11]. Uchino et al. proposed a new classification of PPHA, naming "type 1" the usual PPHA arising from the cervical ICA and "type 2" the PPHA arising from the ECA. There is also a "PPHA variant" when the posterior-inferior cerebellar artery (PICA) arises directly from the carotid system without connection to the VA [12]. PPHA is mostly an incidental finding on angiography and MRI, as was the case in our patient in whom, PPHA was an incidental finding with falx meningioma as a primary finding on MRI performed for chronic headache. To the best of our knowledge and to date, total number of 61 cases of PPHA has been reported in the literature out of which 17 cases were associated with aneurysm and 5 cases associated with arteriovenous malformation [2,10,13,14]. This anomaly is commonly associated with aneurysm and AVM. It is also clinically important when carotid endarterectomy is to be considered. In conclusion, it is important to have the knowledge of this anomaly, and also its association with vascular abnormalities for planning any neurointervention surgery and skull base surgery to prevent possible risks.

Conflict of Interest

The authors declare that they have no conflict of interest.

References