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ANATOMIC VARIATIONS

Bilateral persistent hypoglossal arteries: a case report and literature review

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Abstract
Bilateral persistent hypoglossal arteries (PHAs) are extremely rare, with only 5 cases reported in the English-language literature. Using magnetic resonance angiography, we diagnosed a case in which the left side was a typical PHA and the right side was presumed a PHA variant that supplied only the posterior inferior cerebellar artery.

Keywords Carotid-vertebrobasilar anastomosis · Cerebral arterial variation · Magnetic resonance angiography · Persistent hypoglossal artery · Persistent hypoglossal artery variant

Introduction

A persistent hypoglossal artery (PHA) is the second most common congenital anastomosis between the internal carotid artery (ICA) and vertebrobasilar system (VBS). Their occurrence bilaterally is extremely rare, with only 5 cases, diagnosed using various image modalities, reported in the English-language literature [1, 2, 4, 6, 7]. We diagnosed such a case using magnetic resonance (MR) angiography.

Case report

An 87-year-old woman with medical history that included atrial fibrillation presented with recent fatigue. She demonstrated no neurological symptoms, had an unremarkable neurological examination, and underwent cerebral MR imaging and extra- and intracranial MR angiography using a 3-T scanner to assess for brain infarction and arterial lesion. MR angiography was obtained using a standard three-dimensional time-of-flight technique.

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MR imaging showed no significant abnormality except lesions of chronic ischemic white matter disease, and maximum-intensity-projection images of MR angiography revealed an anomalous artery arising from the cervical ICA on either side (Fig. 1). The left side was dominant and continued to the basilar artery, and the right side was small in caliber with the distal segment not clearly depicted. MR angiographic source images revealed both arterial branches passing through the hypoglossal canals (Fig. 2). Thus, we diagnosed bilateral PHAs. Bilateral vertebral arteries (VAs) were not identified.

Discussion

In early embryonic development, there are 4 types of fetal carotid-vertebrobasilar anastomoses, which include the primitive trigeminal, hypoglossal, otic, and proatlantal intersegmental arteries. As the embryo develops, the posterior communicating arteries develop, and the anastomotic arteries begin to regress at approximately the 30–40th days of fetal development [5].

When this regression fails, the embryonic arteries persist. Usual PHA is the second most common carotid-vertebrobasilar anastomosis and well described by angiography. Its reported prevalence ranges from 0.027% to 0.1% by catheter angiography [11] and is 0.29% by computed tomography (CT) angiography [9]. The PHA usually arises from the cervical ICA, enters the posterior fossa via the hypoglossal canal, and finally anastomoses with the terminal segment

of the VA. Rarely, it arises from the external carotid artery [8, 9].

Bilateral PHAs are extremely rare. To the best of our knowledge, only 5 cases are reported in the English-language literature. Table 1 summarizes those 5 cases and ours. Uchino and associates [10] recently reported a case considered to represent a variant PHA, in which a small PHA supplied only the posterior inferior cerebellar artery and did not connect with the basilar artery. Three of the 6 cases noted, including ours, demonstrated bilateral PHAs with the smaller artery not obviously connected to the basilar artery. Thus, we consider all of these similar cases to represent variants of the PHA as well. Nevertheless, poor visualization of the peripheral segment of the smaller PHA of our patient did not permit definitive confirmation of the artery as a variant. Figure 3 presents a schematic illustration of our patient. Five of the 6 patients demonstrated left PHA dominance, but the small number of cases

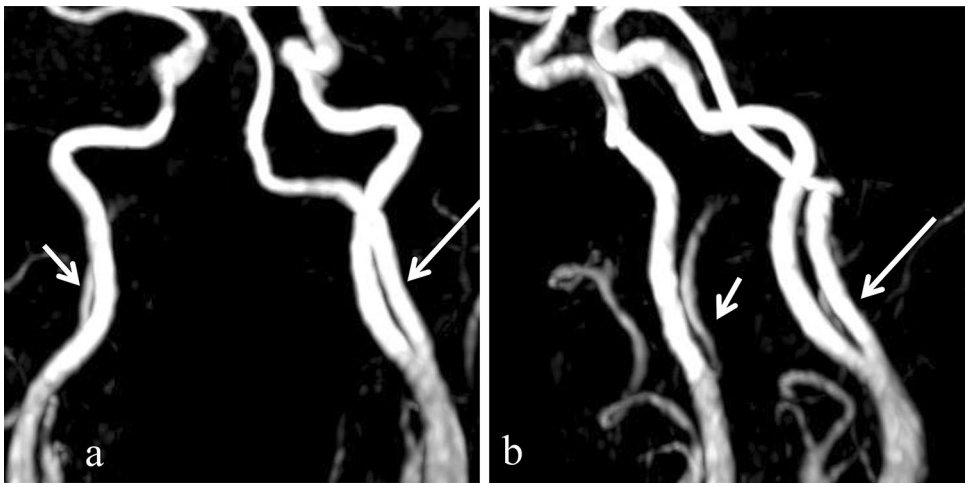

Fig. 1 Antero-posterior (**a**) and left anterior oblique (**b**) projections of magnetic resonance angiography show an anomalous artery arising from the cervical internal carotid artery bilaterally. The left-side artery is large and continues to the basilar artery (*long arrows*); the right-side artery is small, and its distal segment is poorly visualized (*short arrows*). Bilateral vertebral arteries are not identified

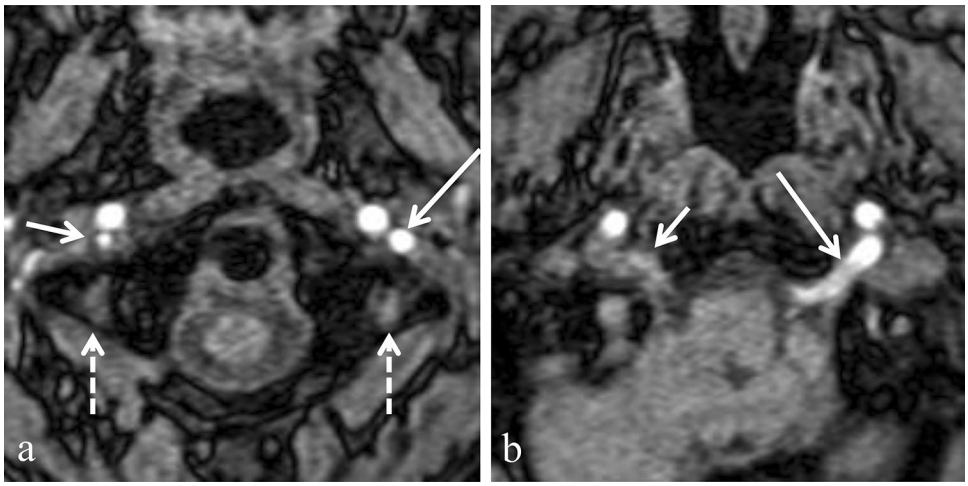

Fig. 2 Magnetic resonance angiographic source images. **a** At the level of the C1 vertebral body, anomalous arteries run along the cervical internal carotid arteries (*long and short arrows*). No arteries are identified in the transverse foramens of the C1 (*dotted arrows*). **b** At the level of the hypoglossal canal, entry of the left large and right small arteries into the posterior fossa via the hypoglossal canals (*long and short arrows*) indicates bilateral persistent hypoglossal arteries

Table 1 Bilateral persistent hypoglossal arteries (PHAs): reported cases in the English-language literature

Reference/year of publication	Age (years)/ gender	Dominant side and possibility of PHA variant in contralateral side	Presented radiological figures
[2] 1976	39/M	Left	Catheter angiography
[4] 1985	59/M	Left, possibly right PHA variant	Catheter angiography
[7] 2012	76/F	Left	MR angiography
[1] 2016	60/F	Left	Catheter angiography, MR angiography
[6] 2017	79/M	Right, possibly left PHA variant	CT angiography, MR angiography
Present case	87/F	Left, possibly right PHA variant	MR angiography

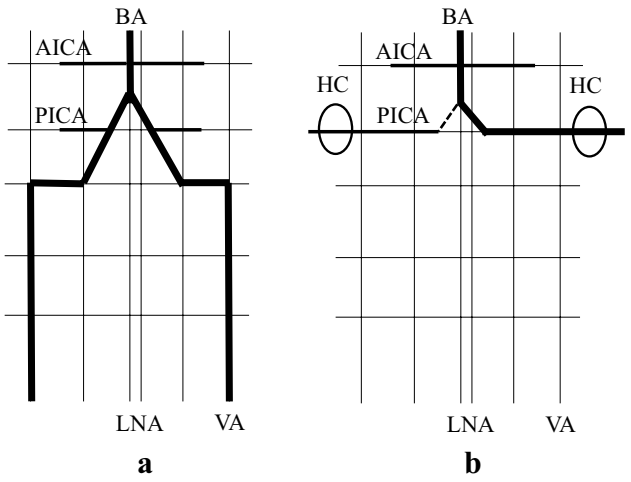

Fig. 3 Schematic illustration of the vertebrobasilar system on anteroposterior projection (modified from Ref. [3], Lasjaunias, Berenstein, ter Brugge). **a** Normal development; **b** presumed development of our case. *AICA* anterior inferior cerebellar artery, *BA* basilar artery, *HC* hypoglossal canal, *LNA* longitudinal neural artery, *PICA* posterior inferior cerebellar artery, *VA* vertebral artery

did not allow us to assess the statistical significance of laterality.

PHAs are usually asymptomatic and have no clinical significance. However, Garge and colleagues [1] reported a case in which PHA with aneurysmal dilation may have caused subarachnoid hemorrhage, and Patira's team [6] suggested that arterial embolization from fetal anastomotic vessels, including PHA, could produce stroke that can mimic a cardiogenic cerebral embolism clinically. In the case of PHAs, bilateral VAs are usually hypoplastic or aplastic [11]. Our patients did not have bilateral VAs. A similar case to our patients with absent VAs diagnosed by MR angiography [7] was previously reported. In cases with brain infarction and acute cerebrovascular disease, therefore, we believe that recognition and accurate identification of PHAs using MR angiography and/or CT angiography could be valuable in evaluating underlying causes and diagnosis.

Conclusion

We presented MR angiographic images of an extremely rare case of bilateral PHAs, in which the smaller artery may represent a variant of the PHA. Careful observation of MR angiography, including source images, may help in recognition and confirmation of such rare arterial variations.

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Author contributions MO and AU designed and carried out the study, acquired data, and drafted and critically reviewed the manuscript. All authors have read and approved the final manuscript.

Compliance with ethical standards

Conflict of interest We declare that we have no conflict of interest.

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