

Persistent primitive hypoglossal artery: an incidental autopsy finding and its significance in clinical practice

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Persistent primitive hypoglossal artery (PPHA) is a recognised, albeit infrequent, intracranial vascular anomaly usually detected during angiography. Its presence is associated with an increased incidence of aneurysm, arteriovenous malformation and ischaemic stroke. A unique case of PPHA discovered during autopsy is described. Additionally, the significance of PPHA in neuroscience is discussed in detail.

Key words: persistent primitive hypoglossal artery, cadaveric finding, cerebrovascular disease

INTRODUCTION

In the 3 mm human embryo the internal carotid artery anastomoses with the basilar artery by means of the primitive trigeminal, otic and hypoglossal arteries from the top downwards. These vascular communications are normally soon obliterated, although in rare cases they persist into adult life and are referred to as persistent anastomotic arteries [22, 24].

The persistent primitive hypoglossal artery (PPHA) is an unusual carotid-basilar communication typically detected coincidentally during cerebral angiography performed for another diagnostic purpose. However, its identification is clinically important during several neurosurgical procedures, since the presence of a PPHA has occasionally been linked to a variety of intracranial vascular pathology [23, 28].

A case of a PPHA identified incidentally during cadaveric preparation is reported and the importance of this unusual vessel in the neurosurgical field is analysed.

CASE REPORT

During regular dissection and after brain exclusion for the purpose of demonstrating the skull base vascular system an extremely rare variation of the vertebrobasilar system was noticed. In particular, an extremely large right PPHA was identified, which coexisted with a hypoplastic left vertebral artery. After performing careful dissection we concluded that the PPHA originated from the posteromedial wall of the lower portion of the cervical part of the right internal carotid artery at the level of the C4 vertebra (Fig. 1).

The persistent primitive hypoglossal artery entered the hypoglossal canal and then emerged from this canal into the posterior cranial fossa. It was directed superiorly and medially and after a distance of approximately 1.5 cm was united with a hypoplastic left vertebral artery at the right side of the lower portion of the clivus. In this way a large basilar artery was formed measuring 4.3 mm in diameter and directed right to the midline of the clivus. It is noteworthy that the ipsilateral vertebral artery



Figure 1. Photograph of the cadaveric specimen demonstrating the large persistent primitive hypoglossal artery (HA) arising from the posteromedial wall of the lower portion of the cervical part of the right internal carotid artery (RICA).

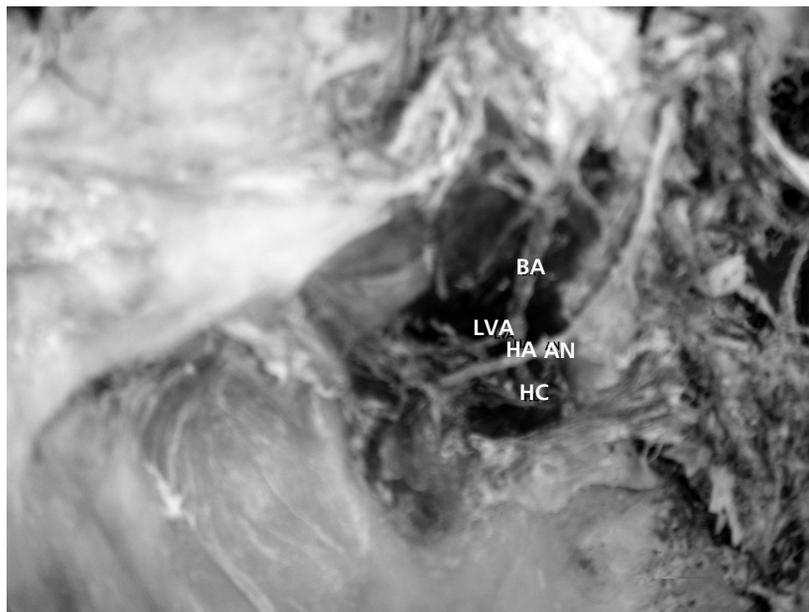


Figure 2. Photograph of the cadaveric specimen showing the persistent primitive hypoglossal artery (HA) emerging from the hypoglossal canal (HC). After a distance of 1.5 cm it is united with the left hypoplastic vertebral artery (LVA), forming a large basilar artery (BA). The abducens nerve (AN) is also visible.

was aplastic and replaced by the right PPHA. Prior to the anastomosis with the left vertebral artery and at a distance of 3 mm PPHA was crossed superoposteriorly by the right abducens nerve. The latter was located at a distance of approximately 5 mm (Fig. 2).

DISCUSSION

During embryological development two cerebral arterial systems can be distinguished, the anterior and the posterior. During the first days of growth (when the embryo is 4–5 mm in length) the posterior system is not

supplied because vertebral arteries have not yet been formed. For this reason the future basilar artery receives blood by means of carotid-basilar anastomoses. In particular, segmental arteries at the level of the brain stem are named after the cranial nerves with which they course (the trigeminal, otic and hypoglossal arteries). These segmental arteries normally regress during later stages of development, although sporadically such an artery may remain into adult life, when in most cases it is identified symptomatically [5, 22].

The persistent primitive hypoglossal artery is a form of segmental artery anomaly characterised by a dorsoventral anastomosis during early foetal life. Although rare, it is the second most common of these embryonic connections, following the persistent trigeminal artery in frequency. It was first described in 1889 by Batujeff incidentally during an autopsy. In 1922 Oertel termed it the "hypoglossal artery" because this artery passes through the hypoglossal canal together with the hypoglossal nerve. Later, in 1950, Lindgreen described PPHA by angiography. Hundreds of PPHA cases have been reported over the years, the vast majority identified during cerebral angiography. The estimated incidence in angiography is between 0.03 and 0.26%, whereas in autopsy studies the percentage remains unknown [1, 6, 9, 10, 13, 16, 23, 30].

The diagnosis and definition of PPHA is grounded on four criteria, according to specific angiographic findings first proposed by Lie and later revised by Brismar:

- 1: the PPHA leaves the internal carotid artery as a large extracranial branch at the level of C1 to C3;
- 2: the PPHA enters the skull through the hypoglossal canal;
- 3: the basilar artery is filled only beyond the point where the artery joins it;
- 4: the posterior communicating arteries are absent or the ipsilateral vertebral artery may be hypoplastic or aplastic.

Brismar revised these criteria in 1976, when he reported a case of persistent hypoglossal artery in which a well-developed posterior communicating artery was found. He suggested that only the first two anatomical criteria 1 and 2 could reliably identify the anomalous hypoglossal artery. The present cadaveric finding fulfilled all the above mentioned criteria except for the fact that it originated at the level of C4 vertebra [3, 14, 15].

Currently segmental arteries such as PPHA can be demonstrated satisfactorily by means of computed tomographic angiography (CTA), magnetic reso-

nance imaging (MRI) and magnetic resonance angiography (MRA). These alternative diagnostic techniques provide excellent anatomical localisation and visualisation of PPHA and all its parts and at present seem adequate for the detection of possible carotid-basilar communications [7, 17, 21].

In the present case PPHA was recognised during an autopsy. The presence of a PPHA, as well that of several vascular anomalies in the surrounding area, was presented very clearly during dissection. Our case exhibits the following important findings:

- 1: the PPHA traverses the clivus to the right of the midline;
- 2: its precommunicated portion (prior to its join with the left vertebral artery) is crossed superoposteriorly by the right abducens nerve at a distance of 3 mm;
- 3: its origin is located at the level of the C4 vertebra instead of the usual C1–C3 level.

It is possible in the case of aneurysm or thrombosis of PPHA for the associated right abducens nerve to be damaged. Since the event was incidental and undisputed, cerebral angiography was not performed. Although the necessary criteria for establishing a diagnosis of a PPHA refer to conventional angiography, they can be applied to our case as well.

In neuroscience, and especially in the field of neurosurgery, the presence of a PPHA is of clinical significance. Since both the anterior and the posterior cerebral circulations are dependent on the internal carotid artery supply, the identification of such a foetal circulatory pattern is important before a decision is taken on how to perform certain surgical or endovascular interventions. It must be emphasised that this persistent artery should not be confused with dilated anastomotic channels that occur after thrombosis of the internal carotid and vertebral arteries and sometimes after thrombosis of the common carotid artery [6, 21, 26].

The persistent primitive hypoglossal artery may be associated with an anomalous structure of the vessel wall and exposes the basilar trunk to unusual haemodynamic stress, leading to a predisposition to the development of aneurysms and arteriovenous malformations. Furthermore, the fragility of the vascular wall and/or haemodynamic stress because of the presence of a PPHA have reportedly been implicated in aneurysm formation. A total of 40 cases of PPHA associated with ruptured aneurysms and 6 with arteriovenous malformations have been described in the literature. The persistent primitive hypoglossal artery is considered to have some association with

aneurysms arising in the posterior circulation. Regarding the operative treatment of aneurysms associated with a PPHA, of the 31 patients who have previously been reported as having undergone surgery 28 underwent aneurysm clip placement [11, 12, 19, 20, 23].

The presence of a PPHA has also been implicated in an increased incidence of atherosclerotic cerebrovascular disease. Since the origin of PPHA from the internal carotid artery forms analogous flow dynamics to the carotid bulb, the development of atherosclerotic plaque might be expected. The plaque may extend from the distal internal carotid artery or as an isolated stenosis near the origin of PPHA. This consideration is of utmost significance in threatening cases of carotid and vertebrobasilar ischaemia (ICA stenosis or ICA endarterectomy), because PPHA usually provides the majority of the blood supply to the posterior circulation. Moreover, the clinical evaluation of a patient with a PPHA and cerebrovascular disease may be confusing as a result of the atypical distribution of cerebral emboli [4, 6, 8, 18, 25, 29].

Both carotid endarterectomy and skull base procedures require adequate preoperative imaging, since the preserved communications between the two vascular systems are often associated with altered patterns of blood flow to the brain. A preoperative angiogram provides distinct assistance. During surgery the hypoglossal as well as the ipsilateral internal carotid artery must be carefully approached and identified. Furthermore, maintenance of adequate cerebral perfusion remains a great challenge, and, since PPHA supports a significant amount of collateral circulation, this has to be preserved. The possible absence of the vertebral blood flow suggests that the entire vertebrobasilar system depends on inflow from ICA. The preservation of the blood supply to the brain becomes more difficult when the lesion operated on (e.g. a skull-base tumour, another growing mass or a vascular lesion) is attached or in close proximity to PPHA [2, 27].

CONCLUSION

The presence of a PPHA is always an interesting finding, either when it is detected during autopsy or in angiography, or when it is identified at the time of surgery. A detailed knowledge of its course is always important in planning and performing certain surgical procedures and manoeuvres.

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