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A rare occurrence of persistent hypoglossal artery and its clinical significance

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Abstract

Persistent Hypoglossal Artery (PHA) is an embryological vascular variation mostly originating from the internal carotid artery. The presence of PHA has been associated with the incidence of some diseases such as cerebral ischemia, atherosclerosis, and aneurysm. Here, a very rare case of PHA that was discovered incidentally by Digital Subtraction Angiography (DSA) in Turkey is reported. Endovascular stenting was not performed for this patient. Also, its clinical importance is discussed.

Key words: persistent hypoglossal artery, digital subtraction angiography, rare variation, vertebrobasilar insufficiency

INTRODUCTION

Presegmental arteries are the carotid-vertebrobasilar anastomoses that supply blood from the internal carotid artery to the vertebrobasilar system in the embryonic period. Four

pairs of presegmental arteries, which are named for neighboring structures, arise from the primitive internal carotid artery: the trigeminal, otic, hypoglossal, and proatlantal intersegmental arteries (Fig. 1). After the posterior communicating arteries develop and the vertebral and basilar arteries join, these temporary collateral vessels disappear at approximately 5 weeks of gestation [8]. The first to regress is the otic artery, followed by the hypoglossal artery, the trigeminal artery, and then the proatlantal intersegmental arteries [7]. Rarely, primitive carotid-vertebrobasilar anastomoses do not disappear and they persist into adult life. Persistent Hypoglossal Artery (PHA) is the second most common anastomosis with an incidence of 0.03-0.09% [9]. It has been reported that PHA is more common in females and on the left side [3]. PHA usually originates from the internal carotid artery at the C1-C3 level and rarely from the external carotid artery.

CASE PRESENTATION

A 57-year-old male patient presented to our clinic with complaints of numbness of the left arm and left leg, impaired balance, and severe dizziness. On neurological examination, the patient showed no motor function loss and his cerebellar tests were normal. The patient reported that dizziness increased when standing up and partially improved after lying down. Doppler Ultrasonography of the carotid vertebral artery showed the presence of PHA on the right side. Upon detecting 70-99% stenosis of the proximal right internal carotid artery and 70% stenosis of the proximal left internal carotid artery along with PHA, imaging with Digital Subtraction Angiography (DSA) was planned. DSA revealed PHA on the right side and 95% stenosis of the PHA, proximal to the right internal carotid artery and distal to the bulb (Fig. 2). The patient also had atherosclerotic stenosis causing 75% stenosis in the proximal left internal carotid artery (Fig. 3). The left vertebral artery appeared hypoplastic compared to the right-sided PHA (Fig. 4). In addition, widespread ischemic gliotic foci were observed in the supratentorial white matter on MR images of the patient (Fig. 5). Endarterectomy was planned for 95% stenosis on the right side of the patient because of anatomical incompatibility. Elective endovascular stenting for 70% stenosis proximal to the left internal carotid artery was scheduled following endarterectomy at our center.

DISCUSSION

Four criteria have been determined for the diagnosis of PHA: 1) the hypoglossal artery originates from the internal carotid artery as a robust branch at the C1-C3 level, 2) enters the posterior cranial fossa through the hypoglossal canal, 3) joins the basilar artery just beyond its origin near the pontomedullary junction, 4) the posterior communicating artery is either absent or not visible on angiography [1]. Vertebral arteries may be hypoplastic or aplastic [7]. It was reported that the diameter of the anterior condylar foramen (ACF) is normally 6-7 mm, which can increase up to 8 mm in the presence of PHA [4].

Begg (1961) first demonstrated the presence of PHA on angiographic imaging [4]. PHA is usually detected incidentally on imaging studies and is often asymptomatic. However, considering the fact that anterior and posterior cerebral circulation depends on the blood supplied by the internal carotid artery, it is crucial to identify such a variation before deciding how to perform surgical or endovascular interventions [6].

Although PHA is considered an anatomical variation of embryological origin, cerebral pathologies should also be considered because any aneurysm may be accompanied by vascular diseases or atherosclerosis [7]. Presence of PHA may be associated with abnormal vessel wall structure. Accordingly, as reported by former studies, exposure of the basilar artery to excessive hemodynamic stress may cause the development of aneurysms and arteriovenous malformations. Cases associated with ruptured aneurysms and arteriovenous malformations have been reported in the literature [6].

PHA has also been associated with atherosclerotic cerebrovascular disease. Atherosclerotic plaque may develop because PHA originates from the internal carotid artery, causing hemodynamics similar to that of the carotid bulb. The presence of a plaque in this region is extremely important in patients with carotid and vertebrobasilar ischemia because PHA generally supplies most of the posterior circulation [6]. Additionally, there are studies in the literature showing the presence of posterior cerebral artery fenestrations and cerebral infarction with PHA [7,10,11]. As a matter of fact, areas of atherosclerosis and cerebral infarction were observed in the present study, which is consistent with the literature data.

Angiography is required before performing carotid endarterectomy and surgical procedures to the skull base region. This is even more important in the absence of the vertebral artery, since in that case the internal carotid artery will be the sole source of cerebral blood supply and it will be difficult to maintain cerebral perfusion during surgery to this region.

Although the incidence of PHA varies in different parts of the world, only 7 cases were reported in Turkey based on our literature search [4,7,8,12,5,2,11]. The fact that so few cases were reported in Turkey makes the present case report even more noteworthy.

CONCLUSIONS

While dizziness has many possible causes, it should be borne in mind that a rare variation such as PHA and vertebrobasilar insufficiency as well as carotid artery stenosis, as emerges in this case, may also cause dizziness. DSA may be considered for the diagnosis of PHA in patients with unexplained dizziness and impaired balance. We believe that this case is representative of a very rare variation and will make a valuable contribution to the literature since there are few cases reported from Turkey.

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This study was conducted in accordance with the ethical standards and the 1964 Declaration of Helsinki and its later amendments. Written informed consent was obtained from the patient.

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Figure 1. Persistent carotid vertebro-basilar anastomoses. Black and green arrows show trigeminal artery, blue arrow otic artery, yellow arrow hypoglossal artery and purple arrow indicates proatlantal artery. Srinivas MR et al. (2016) “Persistent Primitive Hypoglossal Artery (PPHA) - A Rare Anomaly with Literature Review” adapted from his work.

Figure 2. Red arrow shows severe stenosis of the PHA, proximal to the right internal carotid artery, and distal to the bulb on the right side of the patient on DSA imaging; yellow arrow indicates PHA. Green arrow shows common carotid artery, blue arrow external carotid artery and purple arrow proximal right internal carotid artery.

Figure 3. Red arrow shows stenosis of the left internal carotid artery on DSA imaging. Green arrow shows common carotid artery and blue arrow external carotid artery.

Figure 4. Left vertebral artery appears hypoplastic compared to right side PHA. Yellow arrow indicates left vertebral artery, green arrow PHA, blue arrow right internal carotid artery and white arrow left subclavian artery.

Figure 5. Yellow arrows show diffuse hyperintense millimetric ischemic gliotic foci in the supratentorial white matter on brain MRI T2-FLAIR sequence.