CASE REPORT

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Persistent Hypoglossal Artery

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The persistent hypoglossal artery(PHA) is a rare anomaly that belongs to the group of embryonic carotid-basilar artery anastomoses that may occur in adults. The most commonly reported type of such an anastomosis is the primitive trigeminal artery, followed by the PHA. We report a 35-year old man, hospitalized because of an intraventricular hemorrhage, who was found to have a right persistent PHA. Three-dimensional computed tomography(CT) angiography provided excellent anatomical topology of the anomaly. To our knowledge, this patient is the first case of a PHA identified by this means in Korea.

KEY WORDS : Hypoglossal artery · Cerebral angiography · Three dimensional computed tomography angiography.

Introduction

The persistent hypoglossal artery(PHA) is a rare embryonic carotid-basilar artery anastomosis, with a reported incidence of between 0.03% and 0.26% diagnosed by cerebral angiography[3,5,11,13,21]. In Korea, there has been only one published report of the PHA[24]. We report a case of PHA detected by cerebral angiography and subsequently corroborated by three-dimensional computed tomography(CT) angiography.

Case Report

A 35-year old man was admitted to the emergency unit of our hospital with an episode of severe headache, nausea and vomiting. On neurological examination, he was deeply drowsy with neck stiffness. CT scans of the brain revealed an intraventricular hemorrhage. Angiography was performed by Seldinger's percutaneous puncture of the right femoral artery, with selective catheterization of the right and left carotid artery and both vertebral arteries. There was no aneurysm or arteriovenous malformation observed. Selective catheterization of the right carotid artery disclosed a large PHA originating from the right internal carotid artery at the level of the C2 vertebra, which continued as the basilar artery (Fig. 1). The right vetebral artery tapered at the level of the upper portion of the C2 body, and intracranial branches were not visible. Catheterization of the left subclavian artery demonstrated that the left posterior inferior cerebellar artery terminated the left vertebral artery. The left carotid artery appeared normal. Three-dimensional CT angiography readily visualized the PHA as it coursed rostrally along the anterior condyloid foramen (hypoglossal canal; Fig. 2, 3). The patient was underwent extraventricular drainage. His postoperative course was uneventful, and he is being followed up at our outpatient clinic.

Fig. 1. This angiogram of the right internal carotid artery(ICA) demonstrates the persistent hypoglossal artery. This originates from the right ICA at the level of vertebra C2.

Discussion

Carotid-basilar artery anastomoses in Korea

The PHA is a form of segmental artery anomaly, established by a dorsoventral anastomosis during early fetal life. In the

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embryo, this exists between an anterior arterial system, corresponding to the ventral aorta, and a posterior one, located anteriorly in the midline of the neural tube[20]. Segmental arteries at the level of the brainstem are named from the cranial nerves with which they course, and include the trigeminal, otic and hypoglossal arteries[4,22]. Their involution coincides with the development of the posterior communicating artery, which takes place from the 30th to the 40th day of fetal life. Although these segmental arteries normally disappear after the 40th day, sporadically such an artery remains patent, resulting in a carotid-basilar artery anastomosis persisting to adult life. The PHA represents a rare such anastomosis, with a reported incidence between 0.03% and 0.26% on cerebral angiography[3,5,11]. The PHA is the second most frequent anomaly after the persistent trigeminal artery, which makes up the vast majority of persistent primitive connections, whereas the persistent otic and proatlantal intersegmental arteries (type 1, corresponding to the first segmental artery and type 2, corresponding to the second segmental artery) are less frequent. The most common channel is the persistent trigeminal artery in the sellar region. This has been observed in 0.1%0.6% of cerebral angiograms[6,8,17]. Twenty-two people with persistent trigeminal arteries have been reported in Korea[12,15,18,25,30]. Other channels through the otic, hypoglossal, and proatlantal intersegmental arteries are even more rare. The first angiographic demonstration of a PHA was that of Begg[2]. In the following decades, angiographic and neurosurgical cases have been reported. However, only one such case of a PHA has been reported in Korea; no case of an otic artery, and four cases of proatlantal intersegmental arteries have been reported[25,29].

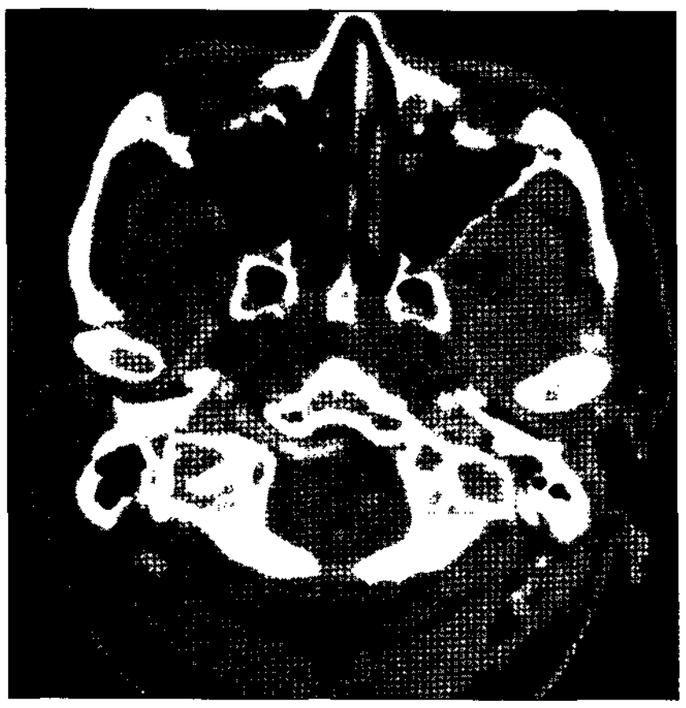

Fig. 2. Contrast-enhanced computed tomography of a bony window, by an axial view, which demonstrates the persistent hypoglossal artery to pass through the right hypoglossal canal.

Fig. 3. Three-dimensional computed tomography angiography demonstrating that the right persistent hypoglossal artery entering the posterior fossa through the right hypoglossal canal.

CT angiography and digital subtraction angiography

Although conventional angiography has been considered the standard for the detection and evaluation of vascular structures, there are alternative diagnostic tools that have different advantages. Magnetic resonance(MR) angiography, MR imaging, and CT angiography have all been used in the diagnosis of PHA[3,9,11,21,23]. CT angiography is a non-invasive alternative that has some advantages. CT angiography affords excellent anatomical localization of this anomaly because of its ability to show the vessel entering the enlarged anterior condyloid foramen, particularly when using three-dimensional reconstruction techniques. Although the diagnosis of PHA has been made most often by using cerebral angiography, such studies are not ideally suited to defining the foramina at the base of the skull. Oelerich reported a detailed demonstration of the vascular anatomy of PHA by the use of MR and CT angiography in 1997[22]. We found that three-dimensional CT angiography readily visualized the PHA as it coursed rostrally along the anterior rim of the foramen magnum after passing through the anterior condyloid foramen. Thus, the technique provides a useful demonstration of anatomical relationships. The carotid-basilar artery anastomosis usually can be differentiated with routine angiographic techniques. However, sometimes there may be confusion about the exact classification. Some pitfalls have been pointed out by Anderson and Sondheimer[1]. The most reliable way to establish the diagnosis of a PHA is to track the vessel's course through the anterior condyloid foramen, which can be enlarged up to 18mm[31]. Three-dimensional CT angiography is thus an excellent tool to prove this anatomical criterion. In our patient, it also demonstrated the course of the PHA through the anterior condyloid foramen. With increasing use of CT angiography in the examination of cerebral vessels, more cases of PHA will be found using these methods.

Clinical implications

A PHA is related to alterations to the anatomy of cerebral circulation. Although it can be observed as an incidental finding by cerebral angiography, its presence has been associated with clinical implications[5]. The persistence of a fetal circulation pattern in a carotid-vertebrobasilar artery anastomosis is of clinical significance in the case of surgical or endovascular intervention. Identification of this anomaly is clinically important before deciding on carotid endarterctomy or surgery to the base of the skull[22]. This is because both the anterior and posterior cerebral circulations depend on the arterial supply of internal carotid artery. The recognition of PHA as a member of the group of persistent carotidbasilar artery connections and the reported incidence of related diseases has led to some pathogenetic considerations. One hypothesis is that the anomaly is related to the formation of atherosclerotic plaques affecting the PHA lumen. Because the origin of the PHA from the ICA forms flow dynamics analogous to those at the carotid bulb, the development of an atherosclerotic plaque can be expected[28]. Another implication is that in cases of atherosclerotic disease, the clinical assessment of a patient with a PHA may lead to some confusion caused by the atypical distribution of cerebral emboli. Although such carotid-basilar anastomoses rarely persist into adulthood, the dual presence of anterior and posterior circulation symptoms should not prevent the clinician from obtaining an angiogram[7]. PHA is also associated with an increased incidence of aneurysms, arteriovenous malformations and atherosclerotic disease[10,11,14,26-28].

The latter may present as a plaque extending from the distal carotid artery into the internal carotid artery(ICA) or as an isolated stenosis near the origin of the PHA. It has been suggested that a PHA may be associated with anomalous structures of the vessel wall and exposes the basilar trunk to unusual hemodynamic stresses predisposing to the onset of aneurysms[11]. The group of persistent primitive arteries, to which PHA belongs, is also implicated in Moyamoya and quasi-Moyamoya disease[13,19]. The detection and investigation of an uncommon vascular anomaly, such as the PHA, by an interventional or a non-interventional approach, is of clinical importance, as its presence is associated with several diseases.

Conclusion

The PHA is a rare persistent embryological connection between the internal carotid and the basilar artery. To our knowledge, this patient represents the first case of a PHA identified by three-dimensional CT angiography in Korea.

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