PERSISTENT CAROTID-VERTEBROBASILAR ANASTOMOSES

How and why differentiating them?

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SUMMARY

The persistent carotid-vertebrobasilar anastomoses (PCVBA) can be explained by an interruption of the vertebrobasilar system (VBS) embryogenesis. We present two very rare cases of persistent anastomoses: a hypoglossal artery and a type I proatlantal artery, insisting on the angiographic criteria allowing differentiation.

After a brief review of the embryogenesis of the VBS, we describe the different types of persistent anastomoses (hypoglossal, type I and II proatlantal, trigeminal and otic arteries). We will insist on the potential risks, not well-known, but typical of each anastomosis. PCVBA usually are incidental findings but imaging follow-up may be required since aneurysms may develop.

Key words: hypoglossal artery, proatlantal artery, trigeminal artery, otic artery, angiography.

Carotid-vertebrobasilar anastomoses usually disappear during embryogenesis. Their persistence in adult patients is sometimes pathological and may result in the development of an aneurysm or a compressive syndrome. The incidence of associated intra-cranial aneurysms is higher in this population. These anastomoses are frequently associated with proximal or distal arterial pathology.

We present two cases of persistent carotid-vertebrobasilar anastomoses (PCVBA): a hypoglossal artery and a type I proatlantal artery, insisting on the radiological criteria which allow to differentiate and distinguish them from the more frequent trigeminal artery and the rare otic artery. We will then describe the potential risks of the different types of anastomoses.

CASE REPORTS

Case n°1 (figure 1)

A 64 year-old man presented with subarachnoid hemorrhage related to a ruptured anterior communicating artery aneurysm. Besides the aneurysm, the pre-treatment angiographic examination showed severe cervical carotid stenosis. The lesion was located at the level of a large common trunk just distal to the carotid bulb. Shortly after its origin, the trunk divided into a cervical internal carotid anteriorly and a second artery of similar diameter posteriorly, which an almost straight course. The posterior inferior cerebellar artery originated from this second artery at the level of the inflection due to its intracranial penetration. This second artery ended into the basilar artery which fed the two posterior cerebral arteries. The two vertebral arteries and posterior communicating arteries were not visible.

Case n°2 (figure 2)

A 58 year-old man presented with a severe asymptomatic ICA stenosis detected at doppler US examination. The angiogram showed that the stenosis was located on the common trunk arising from the carotid bulb and ending in two branches: anteriorly, the cervical internal carotid and posteriorly, an artery of similar size. This last one showed a strong posterior curve before joining the vertebral artery (VA) at the V3 junction. The ipsilateral posterior communicating artery (PCA) is present.
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The ipsilateral VA is not visible. The contralateral VA is hypoplastic.

DISCUSSION

Embryology

In 1954, Padget studied the vascularization on growing embryos. The youngest were 30 days (4-5 mm of diameter) and defined as Padget stade I. Her works enabled her to describe the vertebrobasilar system (VBS) organogenesis [22]. On each side, the embryogenesis lies on the apparition of collateral branches arranging like a ladder, arising from the dorsal aorta (future internal carotid) and generally temporary (figure 3).

At the intra-cranial level, three embryonic arteries participate to the VBS development. They bear the name of the associated cranial nerves: trigeminal, acoustic and hypoglossal. At the cervical level, the first six segmental arteries give rise to the VA. The first segmental artery is called proatlantal artery.

At Padget stade I (30 days, 4-5 mm of diameter) [22], a longitudinal neural artery (LNA) appears on each side. It is cranially supplied by the terminal descending branch of the trigeminal artery and caudally supplied by the ascending branch of the first segmental artery (proatlantal). Laterally, each LNA receives the otic and hypoglossal endings.
At Padget stage II (31 days, 5-6mm of diameter) [22], the two LNA partially merge to form the basilar artery. This last one is collected at the cranial level by the posterior communicating arteries embryologically coming from the internal carotid. At the same time, the ventral trunks of the trigeminal, otic and hypoglossal arteries disappear.

At Padget stage III (33 days, 7-12mm of diameter) [22], one of the caudal and cranial longitudinal collateral branches sets, arising from the first six segmental arteries, fully anastomoses in the caudo-cranial direction. The first segmental artery (proatlantal artery) is linked to the basilar artery by its cranial branch. Its caudal branch is considered as being the starter of the vertebral artery cervical formation in the cranio-caudal direction. When the collateral caudal branch of the 6th segmental artery anastomoses with the cranial branch of the 7th segmental artery (future subclavian artery), the blood flow reverses in the caudo-cranial direction. The ventral trunks of the first six segmental arteries slowly disappear to form the final vertebro-basilar system.

The persistence of one of the first two segmental arteries or one of the three embryonic intra-cranial arteries explains the reported anatomical variations called “PCVBA” in the literature. Probably for hemodynamic reasons, these variations involve a sub-total and/or a total interruption of the vascular embryogenesis [20]. Thus, they are often associated with an agenesis or hypoplasia of the proximal vertebrobasilar system, or abnormalities of the circle of Willis [6]. The persistence of two carotid-vertebral or basilar anastomoses is not rare [1, 18, 19, 31, 33]. Those anastomoses are usually incidental findings [32]. They sometimes are detected in patients with vertebrobasilar ischemic attack secondary to a carotid stenosis, the anastomosis being a pathway for the spread of an embolic agent [3, 13]. They may also compress the surrounding neurological structure, especially if they are large or aneurysmal [1, 6, 11].

The hypoglossal artery (case n°1) (figure 1)

In terms of frequency, the hypoglossal artery is the second carotid-basilar anastomosis (0.02 to 0.09%) after the trigeminal artery [9, 22, 28]. It is bilateral in 1.4% of cases and slightly more frequent in women and on the left side [24]. This anastomosis was discovered by Batujeff in 1889 [4] and Lindgen first angiographically described it in 1950 [17]. It arises from the posterior side of the cervical internal carotid, usually in front of the C1-C2 space but never below the C3-C4 space [6, 24]. It has a short ascending course with a slight medial and posterior curve before penetrating the hypoglossal canal at the skull base. It then ends into the basilar artery. The posterior-inferior cerebellar artery arises from the hypoglossal artery in half of the cases which confirms the embryological link between these two arteries [24]. Indeed, the posterior-inferior cerebellar and ascending pharyngeal arteries are remains of the embryological hypoglossal artery [6, 14].

During vascular imaging, those bone landmarks are the best criteria to identify and prove that the hypoglossal artery penetrates the skull base through the hypoglossal canal [24]. Moreover, standard radiographs in axial views and especially tomodensitometry in bone windows can show widening of the hypoglossal canal. It is a non-specific finding that suggests the possibility of a persistent hypoglossal artery [6].

If the hypoglossal artery persists, the ipsilateral vertebral and posterior communicating arteries are hypoplastic. The contralateral vertebral and posterior communicating arteries are only present in one third of cases [24]. Thus, the posterior portion of the circle of Willis is inconstant. The hypoglossal artery is, then, the exclusive or mainly exclusive feeder of the posterior circulation. Temporarily clamping the hypoglossal artery during carotid endarterectomy presents important ischemic risks [21].

It is frequently an incidental at angiographic evaluation. A hypoglossal nerve (XII) palsy is possible [1] and Kempe describes a case of neuralgia of the glossopharyngeal nerve (IX) irritated by this artery [11]. Reviewing the literature, Resche [24] described six cases of 65 recorded hypoglossal artery aneurysms and 8 cases of associated intra-cranial aneurysms. Probably for hemodynamic reasons, the latter mainly involved the basilar artery proximal to the anastomosis [12]. In four cases, the abnormality was associated with a bone malformation at the cranio-cervical junction [24]. Thus, the hypoglossal artery is the PCVBA most frequently associated with additional pathology.

The proatlantal artery (case n°2) (figure 2)

Forty-four cases of proatlantal arteries are published in the literature [13, 33]. Two types of proatlantal arteries are described. They may arise from the internal carotid (type I Lasjaunias, 18 cases) or from the external carotid (type II Lasjaunias, 26 cases) [14]. The observation we present is the only single French case of type I proatlantal artery published by Bracard in 1984 [6]. Discovered by Gottschau in 1885 [8], the proatlantal artery was first angiographically described by Sutton in 1962 [30]. Only three cases of bilateral proatlantal arteries are described in the literature [18, 19, 33]. This exceptional variation is often discovered incidentally during an angiographic examination.

The type I proatlantal artery arises from the posterior side of the cervical internal carotid, usually in front of the C2-C3 space, rarely at the C4 level. It runs superiorly, posteriorly and medially towards the occipitocervical space. It never runs into the cervical canal. It joins the vertebral artery at the V3 junction in the sub-occipital area before penetrating the foramen magnum [3, 10].

The type II proatlantal artery originates from the external carotid shortly after its origin. Some authors compare its course to the type I proatlantal artery course. For other authors, it joins the vertebral artery before the cervical foramen of C1 through which it travels. Agenesis or hypoplasia of one VÂ is observed in 50% of patients with persistent proatlantal artery [13]. The presence of PCA is rarely specified [13, 19, 33]. No aneurysm is described on its course. Four reports describe associated intra-cranial aneurysms [13].
The only reliable identification criterion is the passage of the proatlantal artery through the foramen magnum at vascular imaging. This course is similar to the vertebral artery course in the suboccipital area.

At the embryologic level, the type I proatlantal artery is a persistence of the first segmental artery arising from the dorsal aorta above the ductus caroticus, future second segment of the cervical internal carotid. Similar to the first cervical nerve, it goes through the occipitocervical space before joining the foramen magnum. On the contrary, the embryological explanation regarding the type II proatlantal artery is more complex and subject to controversy [6, 14]. For some authors, it joins the C1-C2 space before penetrating into the cervical foramen of C1 and the foramen magnum. It could be a persistence of the second segmental artery. A migration of this collateral branch from the dorsal to the ventral aorta during embryogenesis could explain that the type II proatlantal artery arises from the external carotid [14, 15]. The proatlantal artery (type I or II) has close embryologic links with the occipital artery (originating from the first two segmental arteries) [14, 16] and Lui describes the only recorded case of common trunk between those two arteries [19].

The other PCVBA

The persistent trigeminal and otic arteries can easily be recognized because they arise from the intra-cranial internal carotid.

The trigeminal artery is the most frequent PCVBA (0.1 to 0.6% of the angiograms) [2, 6, 26, 28]. In 1844, Quain [23] first described the anatomy and Sutton published the first angiographic observation in 1950 [29]. This artery is very frequently unilateral. It arises from the posterior side of the intracavernous internal carotid, runs towards the petrous pyramid and joins the distal third of the basilar artery. Its sinuous course is typical on a lateral angiogram (figure 4) [2, 6]. In case of a persistent trigeminal artery, the ipsilateral VA and the basilar artery proximal to the anastomosis are often hypoplastic. Classically, the ipsilateral PCA size is inversely proportional to the trigeminal artery size [6, 26]. Its functional role varies and Saltzman distinguishes three types of trigeminal arteries according to the number of injected arteries during a carotid angiogram [27]. This anastomosis is mostly discovered incidentally during angiographic examination or neurosurgery [7]. The most common peripheral neurological symptom is an abducens nerve (VI) irritation in the cavernous sinus leading to diplopia [6, 26]. Facial neuralgias due to the trigeminal nerve irritation are more rare [11, 26]. The frequency of aneurysms on the course of the trigeminal artery or in its distal vascular territory is estimated between 2.8 and 25% [6, 7, 26, 28]. Its association with arterio-venous malformations is subject to discussion [6].

Finally, the persistent otic artery is an exceptional abnormality. Its presence is even controversial [14]. Less than ten cases are reported in the literature [6, 9, 25]. It arises from the internal carotid in the intrapetrous carotid canal, goes out through the internal auditory canal and joins the caudal portion of the basilar artery. The best identifying angiographic criteria is its course through the IAC that can be demonstrated with a Hirtz incidence. The otic artery is usually an incidental finding except in one case reported by Kempe [11] where the acoustic (VIII) and facial nerves (VII) were compressed in the IAC.

How and why differentiating the PCVBA?

The trigeminal, acoustic and type II proatlantal arteries are easily identified by their origins (figure 4 and 5a). On the contrary the level and location of the ostia of the hypoglossal and type I proatlantal arteries cannot be considered as a reliable criteria of differentiation [2]. The main criterion is the point of entrance in the skull base. Digital angiography in lateral projection (figure 2 and 3) can be enough to make a distinction as shown on the lateral diagrams (figure 5b and 5c). The posterior curve of the hypoglossal artery is less important on the lateral views than the strong posterior curve of type I proatlantal artery, due to its entry into the hypoglossal canal anterior to the foramen magnum (figure 1, 2, 5b, 5c) [2].

This distinction is critical since the type I proatlantal artery is far less associated with complications than the hypoglossal artery. No aneurysm has been described on its course and it does not lead to any symptomatic neurological compression. Indeed, it mainly has a contact with the first cervical nerve which potential compression has no clinical manifestation. Moreover, the hypoglossal artery is the most common PCVBA associated with hypoplasia of the two vertebral arteries and the circle of Willis. It is often the exclusive posterior fossa arterial feeder. Therefore, temporary clamping during carotid sur-

**Fig. 4.** – Lateral view of a selective carotid angiogram showing the characteristic sinuous course of a trigeminal artery (arrow).

**Fig. 4.** – Artériographie carotidienne sélective de profil montrant une artère trigéminée avec son aspect sinueux caractéristique (flèche).
Some authors prefer angioplasty which allow shortening the clamping time [5]. The difference of potential complications between these two arteries may be explained by the fact that the proatlantal artery is only a segmental, somatic artery whereas the hypoglossal artery is an embryonic artery mainly for intra-cranial purpose [22].

CONCLUSION

The embryology of the vertebrobasilar system explains the PCVBA. Most are incidental findings and can be easily diagnosed according to their origin. Some of them (the hypoglossal and type I proatlantal arteries) have the same origin. In that case, the analysis of their course on lateral angiographic images should help in making the difference. The hypoglossal artery is more frequently associated with complications than the type I proatlantal artery. The risk of complications, especially the risk of aneurysm, would justify follow-up of these patients using non-invasive vascular imaging (MRA or CTA).

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REFERENCES