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ANATOMIC VARIATIONS

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Persistent carotid-vertebral anastomosis associated with contralateral accessory middle cerebral artery

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Abstract An exceptional case is reported of a complex anomaly of the brain arteries including low left carotid bifurcation, persistent carotid-vertebral anastomosis on the left and accessory middle cerebral artery on the right; the whole posterior circulation was only filled through the anomalous anastomotic vessel which joined with the vertebral artery because of a contralateral hypoplasic vertebral artery and the absence of both posterior communicating arteries. This association has not been previously reported in the literature. The embryological processes leading to these arterial anomalies are discussed. Risks related to a severe carotid stenosis or occlusion and to surgical or endovascular procedures in patients harboring these arterial anomalies are emphasized.

Keywords Vertebral artery · Internal carotid artery · Carotid-vertebral anastomosis · Accessory middle cerebral artery

Introduction

Persistent carotid-vertebral anastomosis is a rare anomaly, resulting in important brain hemodynamic changes, the posterior circulation being fed only by the anomalous vessel. We report a case of persistent carotid-vertebral anastomosis on the left side associated to left low carotid bifurcation and right accessory middle cerebral artery (AMCA). This association has not been described previously.

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Case report

A 14-year-old boy presented with a sudden headache, nausea and slight neck stiffness; at clinical examination the patient was alert with no neurological deficits. A CT scan showed slight bleeding in the perimesencephalic cistern.

A four-vessel angiography through a right transfemoral catheterism was then performed. Common carotid arteries were normal. There was a low left common carotid bifurcation (at C6 vertebral body). The left external carotid artery was normal, with visualization of the ascending pharyngeal and occipital arteries. The left cervical internal carotid artery was significantly larger than the right up to C3 level, where it split into a ventral and dorsal branch, nearly of the same size. The ventral branch run up as the true internal carotid artery, whereas the dorsal branch replaced the left vertebral artery (Fig. 1). This pseudo vertebral artery run straight upwards, nearly parallel to the internal carotid artery, reaching the C3 transverse foramen, where it entered its bony canal and followed the regular course of the vertebral artery, giving origin to the posterior inferior cerebellar artery. A left vertebral artery with subclavian origin was entirely absent. The external carotid was normal, with visualization of the ascending pharyngeal and occipital arteries.

The distal part of the left internal carotid artery and its branches showed a regular pattern (Fig. 2). On selective angiography, the right common carotid artery bifurcated at C3–C4 level. The right internal carotid artery had a regular size and course. In its terminal part an accessory middle cerebral artery (AMCA) (Manelfe type I [1]) was found arising from the internal carotid artery at the angle between the origin of the anterior and middle cerebral arteries (M1 segment). This anomalous

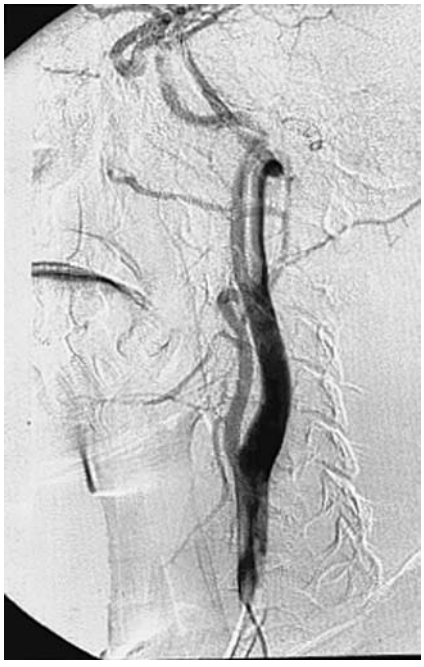

Fig. 1 Angiography of the left common carotid artery in oblique view: low common carotid bifurcation at C6 vertebral body. The internal carotid artery is very large from C6 to C3, where it gives origin to the true internal carotid and vertebral arteries

vessel was slightly larger than the M1 segment of the normal middle cerebral artery and run through the lateral cerebral fissure to reach the insular and parietal cortical regions, giving rise to branches supplying striate and cortical territories. The recurrent artery of Heubner was seen (Fig. 3).

On selective angiography, the right vertebral artery was hypoplastic and ended into the posterior inferior cerebellar artery (Fig. 4). Both posterior communicating arteries were absent. Neither aneurysm or other vascular malformation was seen.

The source of subarachnoid bleed was not found. Treatment with tranexemic acid was started. CT scan at 15 days later showed complete regression of the hemorrage. At present the patient is currently asymptomatic.

Discussion

The type of persistent carotid-vertebral anastomosis described has a phylogenetic explanation. Studies on the vascular trees of several species, from fishes to amphibians, reptiles, birds and mammals, have shown that all these species the internal carotid artery branches into rostral and the caudal trunks. The rostral trunk provides the olfactory system in fishes, amphibians, reptiles and the anterior and middle cerebral arteries in birds and mammals. The caudal trunk provides the tectal and cerebellar arteries in fishes, amphibians and reptiles, the tectal and posterior cerebral arteries in birds and the posterior cerebral artery in mammals. The caudal division becomes the basilar trunk and supplies the entire posterior fossa structures and, in part, of the spinal cord. Thus, in many species the internal carotid artery supplies the whole brain circulation, whereas the vertebral artery is absent [15]. This vascular pattern is also present in some mammals, such as sheep and ox. In higher species, the increasing role of the upper cervical segmental

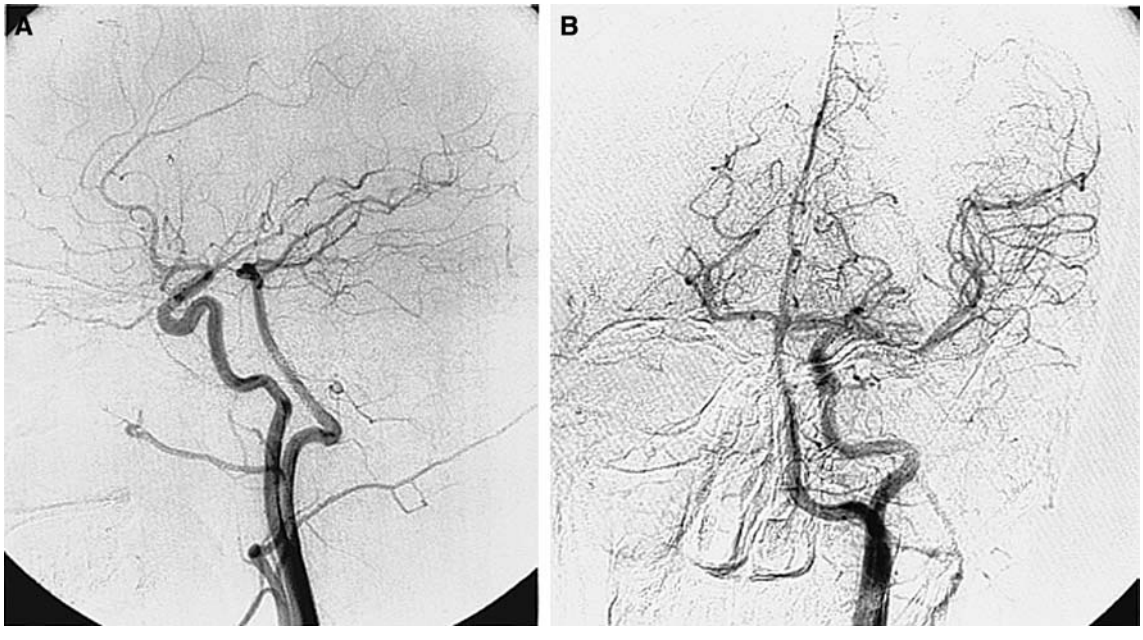

Fig. 2 Left common carotid angiography, in **a** lateral and **b** anteroposterior views: simultaneous injection of the left carotid and vertebro-basilar circulation. Absence of the left posterior communicating artery. Normal external carotid artery with visualization of the ascending pharyngeal and occipital arteries. The posterior inferior cerebellar artery arises from the anomalous vertebral artery

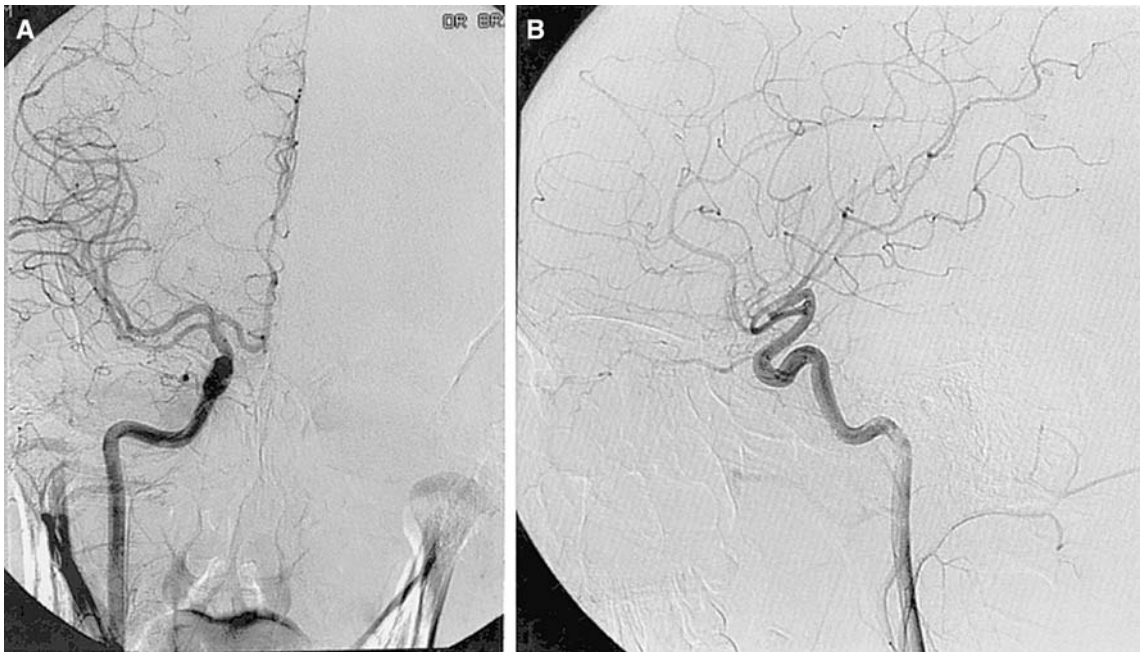

Fig. 3 Right carotid angiography, in **a** anteroposterior and **b** lateral view: presence of an accessory middle cerebral artery, arising from the internal carotid bifurcation-anterior cerebral artery origin. The anomalous vessel is slightly larger than the normal middle cerebral artery and supplies both deep and cortical territories

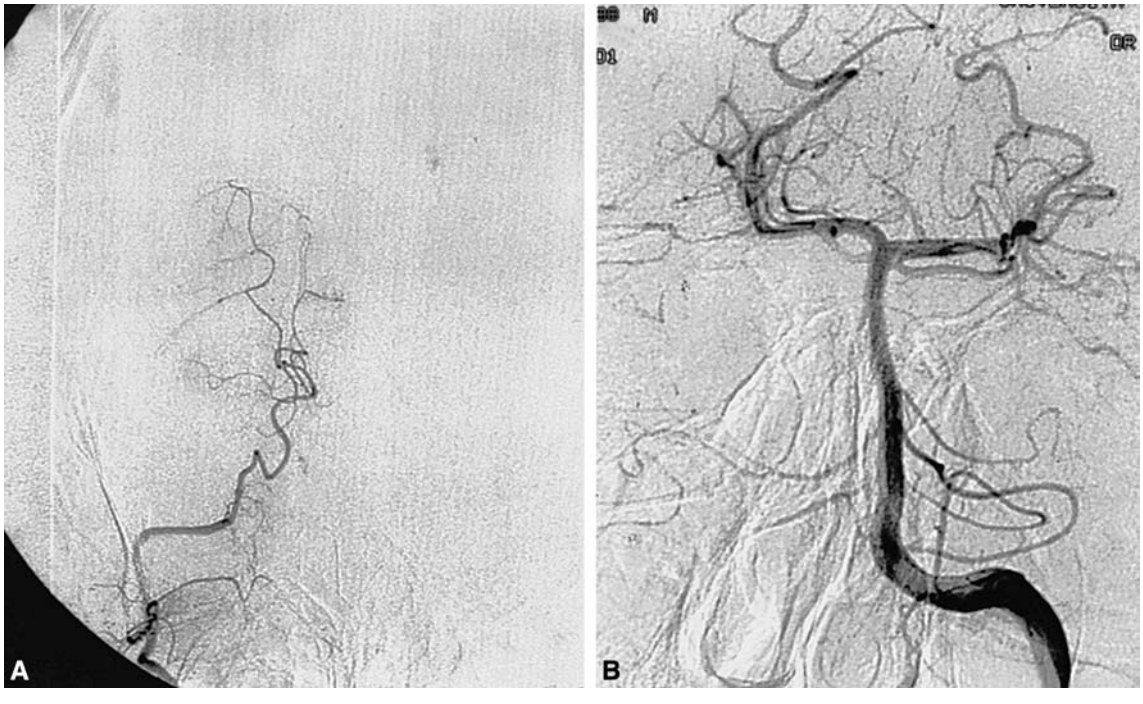

Fig. 4 **a** Selective right vertebral angiography, in anteroposterior view: the artery is severely hypoplasic and ends into the posterior inferior cerebellar artery. **b** Selective angiography of the left vertebral artery through the left carotid artery (anteroposterior view): normal posterior circulation

arteries progressively reverses the direction of flow, so that the posterior cerebral artery origin shifts from the internal carotid artery to the basilar system.

In our case, the vertebro-basilar system is fed by the internal carotid, reflecting the lower species arterial supply.

An anomalous, i.e. non-subclavian origin of the vertebral artery has been observed in 8–10% of humans [2, 6, 20]. Frequently, the vertebral artery may arise from the aortic arch [2, 20]. In rare cases, it may originate from distinct branches of the subclavian artery or brachiocephalic trunk or even from the carotid system arteries. However, a vertebral artery with internal carotid origin seems to be exceptional. Only 14 single cases have been described on angiography [3, 17, 19, 21, 25, 26, 29, 33, 36]. In all cases, the carotid bifurcation was found to be in a regular position, i.e. at the level of either the fourth or fifth cervical vertebra. A pseudo-vertebral artery with internal carotid origin is thougt to be linked to the anomalous persistence of the type I proatlantal intersegmental artery. This artery is defined by its commencement from the internal carotid artery at the level of the second or third cervical vertebra and its oblique upwards and medially direct course. Type I proatlantal intersegmental artery passes anterior to the transverse processes of the cervical vertebrae to reach the gap between the atlas and axis where it enters the vertebral artery canal [3, 17, 26, 36].

However, our case differs from the previous observations for several reasons. First, the bifurcation of the left common carotid artery is more proximal (C6 level). The pseudo-vertebral artery does not correspond to a type I proatlantal intersegmental artery, the ascending course being straight and the artery entering its bony canal formed by the transverse foramina already at C3 level. On angiography, this variation results in an anomalous aspect of the left carotid arterial tree showing two bifurcations, i.e. the bifurcation of the common carotid artery at C6 level and the bifurcation of the enlarged carotid-vertebral trunk into the true internal carotid and pseudo-vertebral artery at C3. This specific angiographic pattern has never been reported.

The low position of the carotid bifurcation at C6 vertebral body or C6–C7 space, with a shorter common carotid, is a rare anatomical variant (0.15% of the angiographic material) [7, 16, 32]. Embryologically, a low carotid bifurcation may be explained by the persistence of the ductus caroticus [4, 22, 24], which may hinder the migration and fusion of the separate origins of the two carotids after the involution of the aortic arch [9, 30]. Thus, this fusion occurs at a lower level [18]. An impairment to common carotid artery growth and elongation by a spur of the carotid bifurcation has also been suggested [13].

The AMCA is also a rare anomaly. It has been reported to occur at a frequency of 0.28% to 2.7% in anatomic studies [5, 8, 12, 23] and in 0.32% of carotid angiographies [35]. According to Crompton [5], any artery running parallel to a regular MCA and supplying its typical vascular region can be defined as an accessory MCA. Teal et al. [31] made a distinction between a duplicated MCA and an accessory MCA. In the first case, an anomalous vessel arises independently from the internal carotid artery, whereas an ''accessory MCA'' is represented by a branch of the anterior cerebral artery (ACA). Abanou et al. [1] used the general term ''accessory MCA'' and, according to the origin of the accessory artery, they subdivided these anomalies into three types. Type I is represented by an artery arising from the internal carotid artery in the angle between the commencements of the ACA and MCA (as found in our case). In type II, the accessory MCA branches off from the ACA proximal to the origin of the anterior communicating artery. In type III the accessory MCA arises from the ACA distal to that point. In most of the cases observed [5, 14], the accessory MCA is smaller than the regular MCA, may end within the sphenoid segment and may partially supply striate territories or even, as in our case, reach the insular and parietal cortical areas.

The middle cerebral artery is a recent phylogenetic acquisition appearing in reptiles. It develops from the lateral olfactory artery of the fish and corresponds to the striate artery of the amphibians [15], continuing its evolution through the mammals and primates. Phylogenetically, both the recurrent artery of Heubner and the middle cerebral artery develop from the group of lateral striate arteries of the anterior cerebral artery; thus, a variable hemodynamic balance exists among these vessels [10]. Takahashi et al. [28] demonstrated that, along with the recurrent artery of Heubner, the accessory MCA represents the primitive vascular anastomosis to the pyriform cortex, with either vessel usually predominating.

Handa et al. [11] suggested that an accessory MCA corresponds to a hypertrophic recurrent artery of Heubner. However, since other investigators [1, 8, 11, 12, 27, 31, 34, 35] detected the coincidence of an independently arising and well recognizable recurrent artery of Heubner and accessory MCA, these two arteries must have separate development. Thus, an accessory MCA corresponds to hypertrophic lenticulostriate arteries sharing a cortical territory of variable size and distribution.

Functionally, the craniocervical arterial pattern found in the our case where the vertebro-basilar circulation depend exclusively on an unilateral vertebral artery with internal carotid origin, may induce severe cerebral circulation impairment similar to that known for other carotido-vertebral and carotido-basilar anastomoses [19, 23]. Due to the absence of both posterior communicating arteries, a severe stenosis or occlusion of the carotid artery proximal to the vertebral artery may result into a dramatic ischemic stroke involving both carotid and vertebro-basilar territories. Furthermore, endovascular procedures performed with the aim of treating carotid stenosis or associated aneurysm are risky. These must be perfomed avoiding transient carotid occlusion in order to preserve the vertebro-basilar flow.

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