

Anterior spinal and bulbar artery supply to the posterior inferior cerebellar artery revealed by a ruptured aneurysm: case report

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The posterior inferior cerebellar artery (PICA) is a vessel located between the intra- and extracranial circulation. The artery is characterized by a complex embryological development and numerous anatomical variants. The authors present a case of the PICA supplied by both a hypertrophic anterior spinal artery and a hypoplastic bulbar artery. This unusual arrangement somehow completes the list of previously published variants, and the spontaneous rupture of a related aneurysm confirmed the fragility of this network. The authors discuss anatomical and treatment considerations.

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THE posterior inferior cerebellar artery (PICA) is a vessel located between the intra- and extracranial circulation and supplies the brainstem but is also in tight connection with the spinal cord. Its origin may be intra- or extracranial, and variations in its course are numerous and often complex. Aneurysms involving the PICA are rare, accounting for only 0.5%–3% of all intracranial aneurysms; the large majority of them (95%–99%) are located at the anterior medullary segment.⁶

Anatomical variations of the PICA can be associated with an increased rate of aneurysmal dilation³ and can also present in atypically distal locations.^{3,7} Treatment in such circumstances may be risky and challenging.^{6,7} We present a rare configuration of the left PICA supplied by a hypertrophic anterior spinal artery (ASA) and a hypoplastic bulbar artery that was revealed by a subarachnoid hemorrhage due to the rupture of a related aneurysm.⁴

Case Report

A 19-year-old man was admitted to our institution with CT evidence of peribulbar subarachnoid hemorrhage revealed by thunderclap headache. Known cardiovascular

risk factors included being overweight (body mass index 27.7) and active smoking.

CT angiography findings were unremarkable, and the patient was referred to the angiography suite for digital subtraction angiography (DSA). The patient's World Federation of Neurosurgical Societies grade was I on admission. A left vertebral artery angiogram revealed the presence of abnormally tortuous vessels at the craniocervical junction.

Superselective microcatheter injections, performed under general anesthesia, revealed an unusual anatomical arterial arrangement with a regular distal PICA supplied by several small branches that originated ventrally from several coronary arteries arising from the ASA and dorsally from a bulbar artery (located at the usual PICA origin, at the level of the hypoglossal nerve), all joining at the PICA posterior medullary segment (also called the tonsillomedullary segment)⁴ (Figs. 1 and 2).

The bulbar branch at its proximal posterior medullary segment harbored a small aneurysm/pseudoaneurysm, consistent with the localization of the bleeding. Because of the high risk of rebleeding, selective exclusion of the aneurysm was considered to be the best option, and, after

ABBREVIATIONS AICA = anterior inferior cerebellar artery; ASA = anterior spinal artery; DSA = digital subtraction angiography; LSA = lateral spinal artery; PICA = posterior inferior cerebellar artery.

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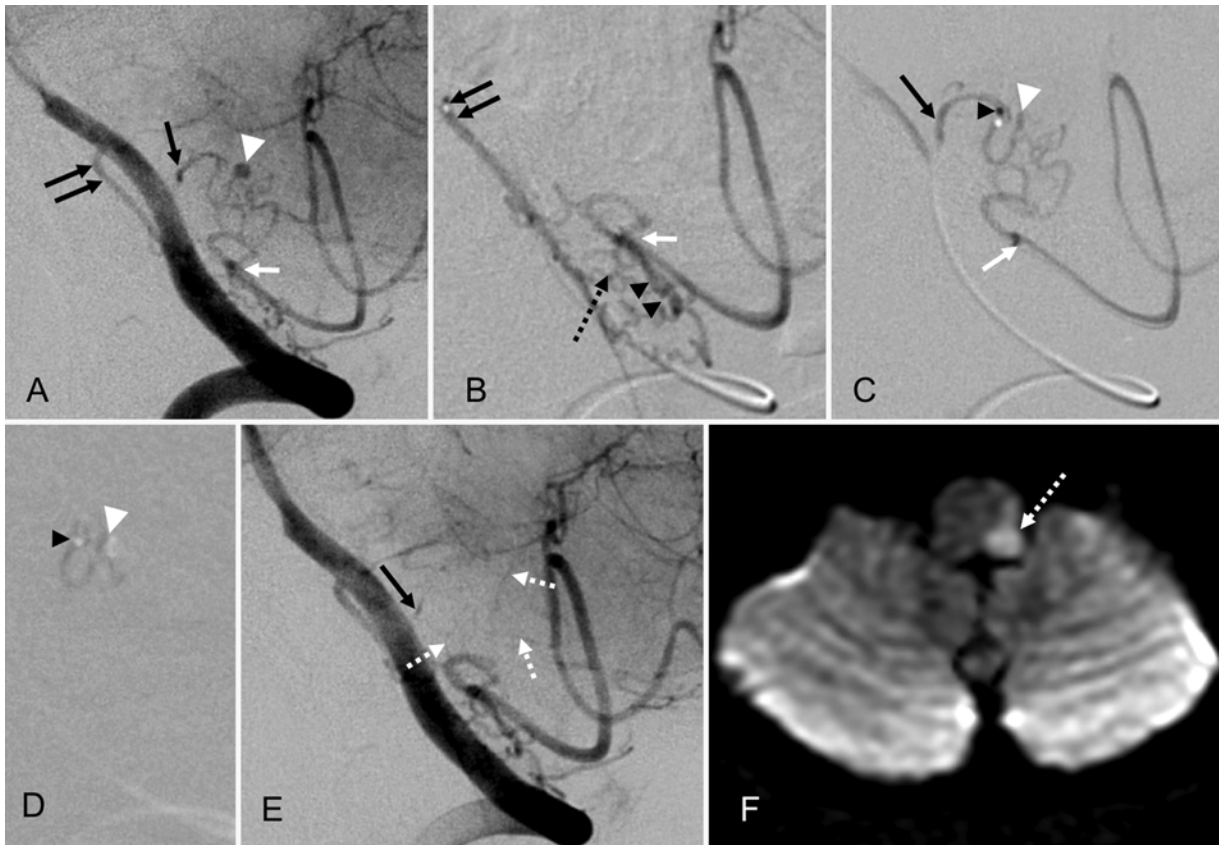


FIG. 1. **A:** Pretreatment left vertebral artery DSA in lateral projection showing the bulbar artery origin (single black arrow), the ASA origin (double black arrows), the aneurysm at the posterior medullary segment (white arrowhead), and the junction between the LSA and the PICA proper (white arrow). **B:** Supraselective injection at the origin of the ASA (double black arrowheads). A hypertrophic vasocorona originates from the ASA. Coronary vessels (dotted black arrow), LSA (double black arrowheads), LSA-PICA junction (white arrow). **C:** Supraselective injection distally into the bulbar artery just before the glue injection, further navigation was not possible; only partial opacification of the aneurysm is seen on DSA due to intrasaccular recumbent stagnating contrast media. Bulbar artery origin (black arrow), aneurysm (white arrowhead), LSA-PICA junction (white arrow), and microcatheter tip (black arrowhead) are shown. **D:** Glue cast after glue injection under blank road map. The removed microcatheter tip (black arrowhead) and excluded aneurysm (white arrowhead) are seen. Note that also the faded white area is part of the glue cast since also the roadmap was affected by the recumbent stagnating contrast media. **E:** Posttreatment left vertebral artery DSA in lateral projection showing the complete occlusion of the aneurysm, a mild spasm at the origin of the bulbar artery (black arrow), and reversed flow toward the glue cast through small anastomotic branches (dotted white arrows). **F:** Postembolization axial diffusion-weighted MR image (3 T) showing hyperintensity of the left dorsal medulla oblongata is seen (dotted white arrow), consistent with an acute ischemic stroke.

multidisciplinary discussion, endovascular treatment was considered to be the most suitable treatment option.

Under general anesthesia, via a 6-F guiding catheter positioned at the distal aspect of the V₂ segment of the left vertebral artery, *n*-butyl cyanoacrylate (Glubran 2, GEM Srl) diluted at 30% in Lipiodol (Guerbet) was selectively delivered through a flow-dependent microcatheter (1.2-F Magic, Balt Extrusion). Distal navigation was obtained with the help of a 0.007-inch micro guidewire (Hybrid, Balt Extrusion) to reach the farthest position. Glue injection was satisfactory, resulting in complete exclusion of the bleeding source with neither proximal nor distal migration of the liquid embolic agent (Fig. 1D). Control angiography revealed patency of all regional vessels except for the embolized segment and mild retrograde flow toward the glue cast (Fig. 1E).

Postprocedure clinical evaluation showed a left Wallenberg syndrome characterized by left Horner's syn-

drome with neither diplopia nor nystagmus swallowing difficulty related to a left ninth cranial nerve palsy, hoarseness and severe hiccups, ipsilateral reduction of pain and temperature sensation of the face, and contralateral loss of pain and temperature sensation of the body. Diffusion-weighted MR images confirmed the presence of a left bulbar stroke (Fig. 1F). The patient was subsequently referred for functional and speech reeducation.

Follow-up at 4 months revealed a general improvement with residual deficit; the main complaint concerned body pain and temperature sensation. The patient was unable to continue his previous occupation and was referred to professional rehabilitation; his modified Rankin Scale score at discharge was evaluated at 2.

Discussion

The PICA is a highly variable artery at the craniocervi-

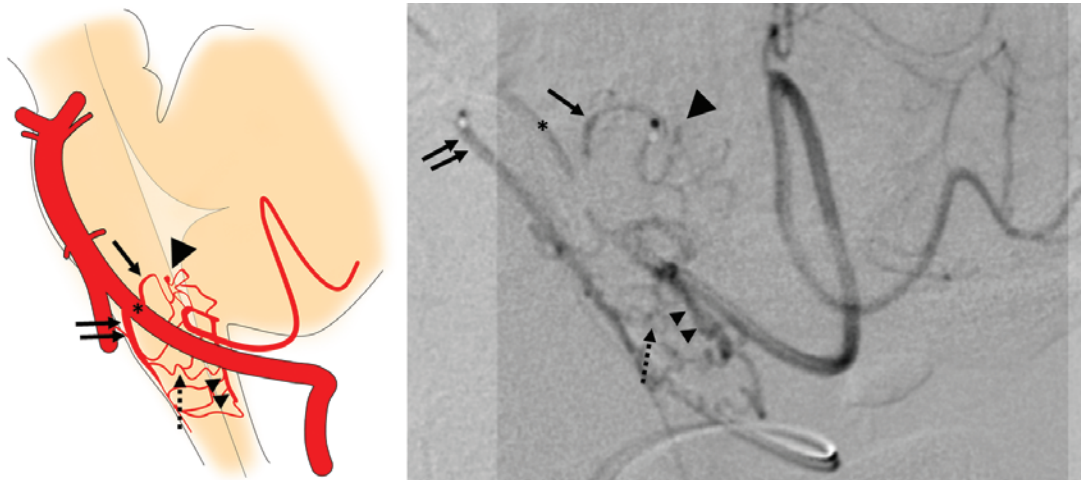


FIG. 2. **Left:** Illustration summarizing the anatomical configuration observed in our case. **Right:** DSA fusion image obtained by the overlap of 2 separate superselective injections at the level of the ASA and of the bulbar artery. The PICA is supplied by the LSA (*double arrowheads*) fed by the ASA (*double arrows*) through the hypertrophic coronary vessels (*dotted arrow*) and by the bulbar artery (*arrow*), which harbors a small aneurysm (*arrowhead*). Only partial filling of the aneurysm is visible. Mild retrograde opacification of the vertebral artery (*asterisk*) is observed. Figure is available in color online only.

cal junction and usually originates from the vertebral artery. It is considered to be the result of the embryological dominance of a posterior radiculopial artery.¹

The PICA usually arises from the dominance of a single pial vessel at the level of the hypoglossal nerve. However, several variations have been reported in the literature, notably a cranial anterior inferior cerebellar artery (AICA)–PICA variant or caudal proatlantal C-1 and C-2 origins characterized by dominance of nearby segmental levels; such cases are usually associated with a hypertrophic bulbar perforator arising at the usual hypoglossal segmental level.

While dominance is certainly the most common anatomical form, up to 2%⁴ of cases may exhibit an unusual codominance, also called a double PICA origin,³ characterized by 2 separate segments supplying the PICA proper. This condition, often underreported, is associated with higher aneurysm prevalence, possibly due to a regional vascular developmental disorganization.³

The PICA is related to the lateral spinal artery (LSA) as much as the posterior radiculomedullary arteries are related to the posterior spinal artery, and this relationship is confirmed by the fact that in most instances (73%) an LSA is visible originating from the PICA itself.¹ Thus, if the PICA proximal to the restiform body (junction of the LSA and PICA)⁵ fails to develop or sufficiently enlarge, usually the LSA becomes the dominant/codominant channel and is supplied either cranially by an anastomotic channel from the AICA or caudally from a segmental branch originating from the extraspinal longitudinal arteries (most commonly the vertebral artery). Interestingly, the location of the PICA's origin seems not only to be related to the LSA but also to affect the origin of the ventral spinal artery.⁵

Indeed, the well-known scheme proposed by Lasjaunias et al.² allows the theoretical possibility for the ASA also to be the origin or co-origin of the PICA, just as the basilar artery regularly gives origin to the AICA.

As demonstrated by this case, the codominant LSA cranially directed to supply the PICA may be supplied by the ASA through the anastomotic network of the coronary vessels. This codominant ASA supply could be either a primitive variant or the result of a postdevelopmental occlusion of a proatlantal feeder followed by hypertrophy of the existing coronary network; the latter seems more likely to explain the features observed in our case (Fig. 3). Indeed, the collateral circulation often develops from embryonic arterial systems, but acquired vascular patterns do not reproduce embryonic stages.¹ In this case, a postdevelopmental arrangement is more likely since multiple irregular tortuous feeders point to the overuse of the adult network rather than the persistence and development of embryonic vessels that usually regress. Nevertheless, this configuration somehow completes the aforementioned scheme and, to our knowledge, has not been previously reported in the literature.

Unfortunately, treatment is still a challenge, since the vessels involved may vascularize eloquent regions of the brainstem and the treatment consists of a selective vascular sacrifice. It is a common opinion that such selective segmental occlusion is usually well tolerated, even if perforators arise next to or from the trapped segment;⁷ nevertheless, the risk is present and should be balanced with the expected benefits.

Alternatively, selective exclusion of the aneurysm(s) plus a vascular PICA-PICA bypass distal to the origin of the aneurysm to protect the medulla via retrograde flow in case of feeder sacrifice has previously been reported at least once.⁶ In our case, this option did not seem to add sufficient benefit since the ASA-PICA network already constitutes a natural bypass, as demonstrated by the postoperative patency of all vessels including a retrograde flow toward the glue cast. A distal bypass nevertheless could have a hemodynamic impact reducing the flow through the proximal network and the shear stress on the wall of its arteries.

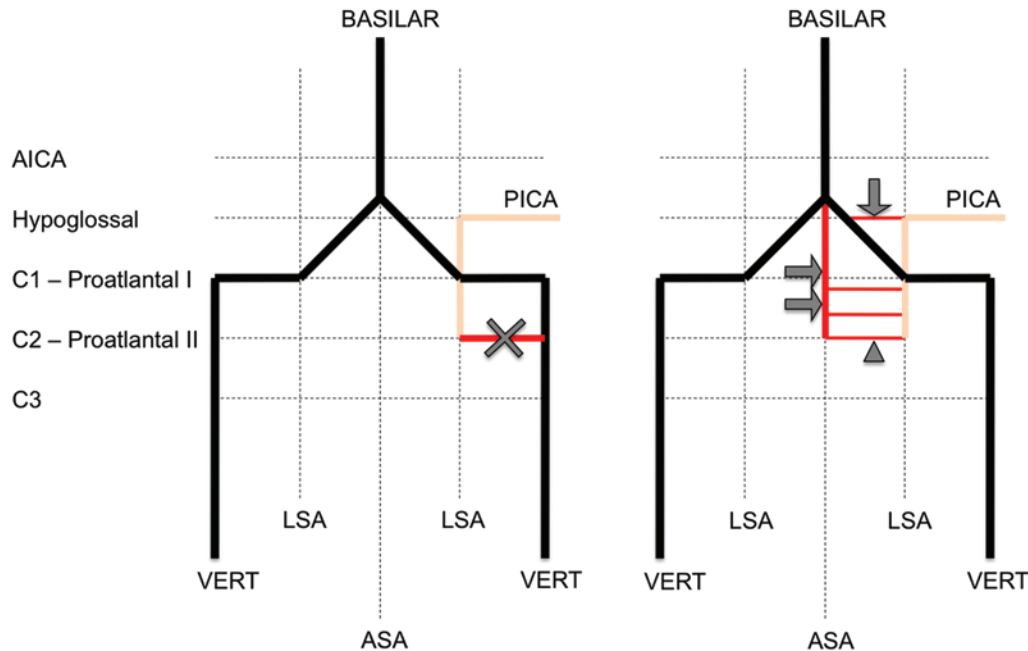


FIG. 3. Schematic of the developmental pathways allowed at the craniocervical junction, depicted in the style shown in Lasjaunias et al.: *J Neurosurg* 63:235–241, 1985. **Left:** A hypothetical insult (X) at the level of a low PICA origin. **Right:** Postdevelopmental hypertrophy of the ASA (double arrows), vasocorona (arrowhead), and bulbar artery (arrow). VERT = vertebral artery. Figure is available in color online only.

This case highlights a particular anatomical arrangement, its associated aneurysmal lesion, and once again⁷ stresses the importance of the medullary perforating vessels, especially in cases of variations of the PICA origin.

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Disclosures

Dr. Sourour reports that he is a consultant and proctor for Covidien and Stryker.

Author Contributions

Conception and design: Gabrieli. Acquisition of data: Sourour. Analysis and interpretation of data: Gabrieli, Clarençon. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors.

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