Subarachnoid hemorrhage caused by posterior inferior cerebellar artery aneurysm with an anomalous course of the atlantoaxial segment of the vertebral artery

Case report and review of literature

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Various anatomical courses of the vertebral artery (VA) and posterior inferior cerebellar artery (PICA) have been described. The authors present a unique case of a subarachnoid hemorrhage resulting from an aneurysm in a patient with an anatomical variation of the extracranial portion of the VA and cervical origin of the PICA. The surgical implications of this variant are discussed, and the pertinent literature reviewed. Subarachnoid hemorrhage caused by rupture of a PICA aneurysm is reported for the first time in association with a rare variation of the course of the VA.

KEY WORDS • vertebral artery • posterior inferior cerebellar artery aneurysm • subarachnoid hemorrhage

POSTERIOR inferior cerebellar artery aneurysms represent a small percentage (0.5–3%) of intracranial lesions. Although relatively rare, numerous anatomical variations have been described for the VA and PICA. We present a unique case in which the atlantoaxial portion of the VA takes an anomalous course in association with an aneurysm arising from the right PICA. After exiting the foramen transversarium of C-2, rather than taking the typical course through the foramen transversarium of C-1, the artery coursed medially between C-1 and C-2 where it entered the posterolateral aspect of the dura of the cervical spine. Similar variations in the course of the VA have been described but to our knowledge have not been associated with an aneurysm. We discuss the relevant anatomy and surgical implications and review the pertinent literature. We also speculate on the association of this anatomical variation with the concurrent right PICA aneurysm.

Case Report

History. This 63-year-old right-handed woman with a medical history significant for coronary artery disease, myocardial infarction, stroke, chronic obstructive pulmonary disease, and cigarette smoking (120 pack years) presented to an outside hospital after becoming unconscious in her car following a brief period of slurred speech and seizures. She had been having moderate-to-severe headaches for 1 month before this event. After endotracheal intubation and stabilization, a head CT scan was obtained, revealing diffuse SAH and intraventricular hemorrhage in the fourth ventricle. She was then transferred to Barnes-Jewish Hospital for further care.

Examination. On arrival to the hospital, physical examination revealed that the patient opened her eyes only to noxious stimuli and did not follow commands. Her pupils were 3 mm in diameter, round, and reactive to light. Her oculcephalic response was normal, and gag and cough reflexes were intact. She was spontaneously moving her upper extremities and weakly moving her legs. Her Glasgow Coma Scale score was 7T (eye opening = 2, verbal = T [intubated], and motor = 5), and she had a Hunt and Hess Grade IV and Fisher Grade IV SAH. A repeated head CT scan was obtained to evaluate for hydrocephalus. A comparison with the previous scan demonstrated ventricular enlargement consistent with acute hydrocephalus and unchanged SAH and intraventricular hemorrhage. A right frontal external ventricular drain was placed. Cerebral angiography studies revealed a multilobulated 6 × 6-mm aneurysm extending medially from the origin of the right PICA below the foramen magnum (Fig. 1).
No osseous anomaly was identified in our case. They are often associated with distal duplications, hypoplasty, and fenestration.

We describe a PICA aneurysm. After a thorough search of Medline and other sources, we have concluded that a VA–PICA complex is not uncommon. Anomalies of the VA–PICA complex are not uncommon. Anomalies of the VA–PICA complex have also been reported.

Although some of these anomalies have been associated with lesions of the PICA, they are often associated with distal aneurysms and are not usually VA–PICA junction aneurysms. Extradural origins of the PICA with and without aneurysms have been reported. An intradural cervical origin of PICA aneurysms has also been described.

Preoperative angiography and intraoperative surgical findings displayed an anomalous course of the VA. The artery did not pass through the foramen transversarium of C-1. Instead, after exiting the foramen transversarium of C-2, the artery coursed medially between the laminae of C-1 and C-2 and then passed into the dura at the posterolateral aspect of the spinal canal between C-1 and C-2 (Fig. 2). The PICA and the associated aneurysm originated in an intradural fashion in the cervical region below the foramen magnum just distal to the site of the VA entry into the spinal canal. The patient’s case was reviewed by the neurosurgical and neurointerventional radiological teams, who determined that the aneurysm was not optimally amenable to endovascular coil insertion given the apparent origin of the PICA from the neck of the aneurysm. Although the patient had multiple medical comorbidities, the anesthesiologists and cardiologists who evaluated her believed that the risk of surgery was acceptable.

Surgery. Clips were applied to the aneurysm through a small suboccipital craniotomy with partial laminectomies of C-1 and C-2 by using a microsurgical technique (Fig. 2 inset and Fig. 3). Intraoperative and postoperative angiography studies exhibited occlusion of the aneurysm and preserved flow through the parent vessels (Fig. 4). The patient’s neurological recovery was good (Glasgow Outcome Scale Score 4).

Discussion

In a review of 300 VA angiograms, Tokuda, et al., reported on the incidence of VA anomalies in 2 to 3% of patients, and such anomalies have been reported in association with osseous anomalies such as Klippel–Feil syndrome. No osseous anomaly was identified in our case. Anomalies of the VA–PICA complex are not uncommon. Lister, et al., examined PICA anatomy in 25 cadaveric heads (50 sides) and noted the presence of a VA in 49 of 50 cerebellar hemispheres and the presence of a PICA in 42. Seven of the 42 PICAs arose from below the foramen magnum, but none had VA anomalies. In a cadaveric study, an extradural origin of the PICA was reported to occur 5 to 20% of the time. Duplications, hypoplasia, and fenestrations of the PICA have also been reported.

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Dr. Sharma and colleagues described a patient without an aneurysm but with anatomy similar to that in our patient, with the course of the VA passing beneath the posterior arch of C-1 in the context of C-2 nerve root compression and occipital neuralgia. These authors reported that only six cases of the VA passing beneath the posterior arch had been documented in the literature. A thorough search of Medline and other sources, we have concluded that a VA anomaly of the type featured in the present study has not been previously reported in association with a ruptured VA–PICA aneurysm.

The PICA is generally divided into five segments, with aneurysms most commonly arising from the proximal anterior medullary segment. We describe a PICA aneurysm that arises from what is ordinarily termed the “anterior medullary segment at the origin of the PICA from the VA.” In the present case, however, the nomenclature is somewhat misleading because the aneurysm is adjacent to the upper cervical spinal cord rather than the medulla oblongata. Aneurysms of the PICA are uncommon but can be potentially dangerous because, as in the case featured here, they can rupture even at relatively smaller sizes. The hemodynamic stress theory posits that aneurysms tend to form in areas of high or abnormal hemodynamic forces (for example, branch points or hairpin turns). Thus, vascular

Fig. 1. Preoperative cerebral angiograms. In the later projection of the right VA (A), an approximately 6-mm lobulated PICA aneurysm is made visible. Because the bone anatomy is well demonstrated, one can appreciate the extracranial position of the PICA aneurysm and its relationship to the dens. Compare the course of the right VA (A) to the left VA in the anteroposterior projection (B). The VA traverses inferomedially after coursing through the foramen transversarium of C-2. Instead of taking a normal course (B), it makes a hairpin turn and passes into the dura between C-1 and C-2. The aneurysm is not as easily seen on the lateral projections (C, right projection; D, left projection), but these projections highlight the abnormal anatomy of the right VA in relationship to the foramen transversarium of C-1 (arrow in C). Digital subtraction angiogram (inset, i), right VA injection, clearly demonstrating the multilobulated PICA aneurysm with its relatively narrow neck.
Posterior inferior cerebellar artery aneurysm with anomalous VA

Fig. 2. Artist’s rendering of the anatomy encountered in the patient in the present case. One can see the inferomedial course of the right VA as it passes beneath the lamina of C-1 and not through its foramen transversarium. The VA then passes into the dura between C-1 and C-2. The left VA takes its normal course through the C-1 foramen transversarium. The PICA aneurysm is intradural but extracranial, located in the cervical region. The inset represents a view after partial C-1 and C-2 laminectomies were performed.

Fig. 3. Intraoperative photograph illustrating the initial surgical exposure. Note the origin of the PICA and the aneurysm in the cervical spinal canal (arrow) and the distal course of the PICA (arrowhead) from right lateral to medial directions over the dorsum of the spinal cord. C = cerebellum; I = inferior; L = patient’s left; R = patient’s right; S = superior.
anomalies such as azygous vessels or fenestrations are commonly found in association with an aneurysm. As discussed, PICA aneurysms often occur in the context of vascular anomalies. It is possible that the unusual course of the right VA created hemodynamic stresses that predisposed our patient to aneurysm formation.

The highlighted case also demonstrates the importance of obtaining a high-quality unsubtracted angiogram so that the location of the aneurysm can be correlated with the bone anatomy. The relationship of the aneurysm to the foramen magnum as well as C-1 and C-2 could have been easily overlooked without the unsubtracted views.

Note that the aneurysm was approached through a small suboccipital craniotomy. Partial laminectomies of C-1 and C-2 and definitive microsurgical clip application were performed. Special care was taken when obtaining proximal and distal control and dissecting the dome of the aneurysm. While achieving proximal control, we encountered the right VA earlier and more medially than normally expected. Anomalies such as this should be considered when surgery is performed in this region, whether it is for an aneurysm or other posterior procedures of the occipitocervical or cervical regions.

Conclusions

An SAH caused by the rupture of a PICA aneurysm in association with a rare variation in the course of the VA was reported for the first time. The unique course of the VA may have contributed to the pathophysiological formation of the PICA aneurysm. The neurosurgeon must remember such anomalies when applying clips to an aneurysm or performing other surgical procedures in this anatomical region.

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Fig. 4. Intraoperative cerebral angiograms in the anteroposterior (A) and oblique (B) projections. The right VA–PICA aneurysm is obliterated by aneurysm clips, and the right PICA is preserved with normal blood flow. Note the position of the clips below the foramen magnum.
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