Multiple Aneurysms on Inter-PICA Communicating Collaterals: Case Report on a Rare Entity

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Abstract

Aneurysms arising from the posterior inferior cerebellar artery (PICA) to PICA communicating collaterals are very rare. Only five cases have so far been described in the literature. Here, we report the first case with multiple aneurysms occurring on separate inter-PICA communicating collateral arteries, where one of the PICAs was occluded proximally and reconstituted distally through the inter-PICA collateral arteries, in a patient presenting with subarachnoid hemorrhage. After standard management, the ruptured aneurysm was resected, and the other unruptured aneurysm was clipped. Side-to-side PICA-to-PICA anastomosis distal to the communicating collaterals was performed in order to maintain collateral circulation to the brainstem. Clinicians should be aware of the possibility of these rare aneurysms, and should consider in situ bypass in the management of PICA aneurysms where the aneurysm is proximal to the telovelotonsillary segment of PICA.

Categories: Neurosurgery
Keywords: aneurysm, posterior inferior cerebellar artery, subarachnoid hemorrhage, arterial anastomosis

Introduction

The posterior inferior cerebellar artery (PICA) is a tortuous artery with extensive anatomical variation [1-2]. PICA aneurysms are rare, representing 0.49-3% of all intracranial aneurysms [3-4]. The majority of PICA aneurysms develop at the anterior medullary segment at the junction of the PICA and the vertebral artery (VA) [3, 5]. Zero point nine to 5.2% of all PICA aneurysms arise distal to the anterior medullary segment [4-6]. Most of the aneurysms develop proximal to the telovelotonsillary segment [5, 5]. Aneurysms arising from small perforating arteries of PICA are exceptionally rare, with only eight cases described in the literature [4, 7-18]. PICA-to-PICA “communicating collateral” aneurysms are even rarer, with only five cases reported [4, 7-10].

Here, we report a case of a ruptured aneurysm arising from an inter-PICA communicating collateral artery at the telovelomedullary segment, along with a similar but smaller unruptured aneurysm from an adjacent inter-PICA communicating collateral. This case is unique in several respects. To the authors’ knowledge, it is the only case reported with one aneurysm on each of multiple inter-PICA communicating collaterals. Furthermore, it is also the only reported case where side-to-side PICA-to-PICA bypass was performed beyond the communicating collaterals in an attempt to increase the circulation on the side of the proximally occluded PICA.
A 76-year-old right-handed Caucasian female with history of hypertension presented with acute onset of severe headache. She was intubated at the local emergency room and was transferred to our facility for further management. Initial CT head demonstrated significant subarachnoid hemorrhage with a bleeding pattern typical of lower posterior circulation hemorrhage (Figure 1A-B).

A brainstem arteriovenous malformation (AVM) with associated aneurysms was initially suspected. On exam, the patient was stuporous. She withdrew her limbs to pain, and cranial nerves were grossly intact. She was classified as Hunt and Hess Grade 4 and Fisher Grade IV. After the patient was stabilized in the neuro intensive care unit, cerebrospinal fluid (CSF) was diverted through ventriculostomy and proper work-up, including MRI brain, CT-angiogram, and catheter angiography, was performed. Catheter angiogram demonstrated that the left PICA was occluded at its origin. It was reconstituted by collateral supply from the right PICA through two communicating collateral arteries, each with one aneurysm, at the telovelomedullary segment (Figure 1C-E). The aneurysm on the more proximal collateral was larger and assumed to have ruptured.
Operation and Postoperative Course

The patient was placed in prone position and underwent a midline suboccipital craniectomy and C1-laminectomy. A serpiginous right PICA, adjacent to a relatively straight left PICA beyond the communicating collateral arteries, was found. The left PICA was occluded at its origin. There was a 6.8 mm ruptured fusiform aneurysm on the more proximal collateral artery and a 5.2 mm unruptured aneurysm on the adjacent distal collateral artery. The larger fusiform aneurysm was resected. In order to maintain collateral flow to the left PICA, a side-to-side PICA-to-PICA anastomosis was performed. The smaller unruptured aneurysm was clipped, leaving a portion of its neck intact.

VIDEO 1: Operative clip showing highlights of the operative technique for inter-PICA communicating collateral aneurysm clipping and side-to-side PICA-to-PICA bypass.

View video here: https://www.youtube.com/watch?v=pA7Ms84GjX4

Intraoperative indocyanine green (ICG) video-angiography showed immediate patency of the bypass and postoperative 3D-rotational angiography confirmed sustained flow through the graft and no further filling of the remaining aneurysm neck (Figure 2A-B).
The patient ultimately required permanent CSF diversion and therefore underwent ventriculoperitoneal shunting with a programmable strate-valve set at 0.5. She also underwent tracheostomy. At one month follow-up, the patient was alert, oriented, with intact cranial nerve function, and following commands in all four extremities, though with slight weakness in both upper extremities. She was also without any sign of pseudomeningoele. She was transferred to a skilled nursing facility for further rehabilitation and eventually discharged. At three months, the patient returned with confusion and ventriculomegaly, and she underwent shunt-revision showing distal obstruction (Figure 2C-D). The valve was changed to a low-pressure valve. Postoperatively the patient did well. At discharge she was alert and oriented, could speak full sentences, and moved all four extremities on command, although she remained marginally weak in both upper extremities.

**Discussion**

PICA-to-PICA communicating collateral artery aneurysms are extremely rare, with only five cases previously reported in the literature.
<table>
<thead>
<tr>
<th>Authors</th>
<th>Age</th>
<th>Sex</th>
<th>Hunt Hess Grade</th>
<th>CT Findings</th>
<th>Aneurysm(s) Diameter (mm); Shape</th>
<th>Variation</th>
<th>Surgery</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hlavin et al. (1991)</td>
<td>45</td>
<td>F</td>
<td>3</td>
<td>Isolated blood in the posterior horn of the left lateral ventricle and mild ventriculomegaly</td>
<td>&lt; 5; saccular</td>
<td>Absent right PICA and AICA with the left PICA supplying through an inter-PICA collateral</td>
<td>Neck clipping</td>
<td>Excellent</td>
</tr>
<tr>
<td>Udono et al. (1997)</td>
<td>51</td>
<td>F</td>
<td>3</td>
<td>Thick hematoma in the cisterna magna, diffuse SAH, and intraventricular hematoma</td>
<td>2.5; saccular</td>
<td>None reported</td>
<td>Neck clipping</td>
<td>Excellent</td>
</tr>
<tr>
<td>Fujiwara et al. (1999)</td>
<td>61</td>
<td>F</td>
<td>3</td>
<td>Hematoma in the third, fourth, and lateral ventricles, and a mild posterior fossa SAH</td>
<td>3; saccular</td>
<td>Absent left PICA with the right PICA supplying through an inter-PICA collateral</td>
<td>Neck clipping</td>
<td>Excellent</td>
</tr>
<tr>
<td>Okuno and Onnishi (2000)</td>
<td>68</td>
<td>M</td>
<td>4</td>
<td>Thick SAH around the medulla and massive fourth-ventricular bleeding</td>
<td>3; fusiform</td>
<td>Absent right PICA with the left PICA supplying through an inter-PICA collateral</td>
<td>Trapping</td>
<td>Poor</td>
</tr>
<tr>
<td>Sugiyama et al. (2012)</td>
<td>60</td>
<td>M</td>
<td>2</td>
<td>SAH with intraventricular hematoma in the third, fourth, and bilateral lateral ventricles</td>
<td>2.5; fusiform</td>
<td>Absent left VA, AICA, and PICA with right PICA supplying through an right inter-PICA collateral</td>
<td>Trapping</td>
<td>Excellent</td>
</tr>
<tr>
<td>Present case</td>
<td>76</td>
<td>F</td>
<td>4</td>
<td>Significant SAH with a bleeding pattern typical of lower posterior circulation hemorrhage</td>
<td>6.8 and 5.2; saccular</td>
<td>Occluded left PICA with communication to the right PICA through inter-PICA collaterals</td>
<td>Resection, neck clipping, and in situ anastomosis</td>
<td>Excellent</td>
</tr>
</tbody>
</table>

**TABLE 1: Table 1: Summary of 6 cases (including the present case) of posterior inferior cerebellar artery inter-communicating collateral artery aneurysms**

SAH = subarachnoid hemorrhage, PICA = posterior inferior cerebellar artery, VA = vertebral artery, AICA = anterior inferior cerebellar artery

Due to the radiologic appearance, the lesion was first suspected to be an AVM with two associated aneurysms. However, catheter angiogram confirmed the presence of two isolated saccular aneurysms on adjacent inter-PICA communicating collaterals originating between the right and left telovelomedullary segments.

PICA aneurysms distal to the anterior medullary segment are frequently associated with variants in circulation [3-4, 8]. All but one of the previously reported cases of inter-PICA collateral artery aneurysm involved congenital contralateral vascular hypoplasia that contributed to aneurysm development secondary to increased flow through the small collateral. A single absent PICA has so far been reported to be the most common vascular abnormality [4, 8-9]. One case described an absent left vertebral artery (VA), anterior inferior cerebellar artery (AICA), and PICA with contralateral supply through the inter-PICA communicating collateral; the right AICA was also connected to PICA and supplied its territory through this collateral artery [7]. Congenital variations like these are assumed to cause the development of collateral arteries enabling ‘physiological’ bypass of the abnormality and perfusion of the contralateral side. However, increased hemodynamic stress and turbulent flow through these small arteries is what presumably leads to aneurysm pathogenesis [6, 8].
All but two inter-PICA collateral aneurysms so far reported have been saccular. All but one developed on a straight portion of the collateral vessel as opposed to a branching point. It is unclear how branchless artery aneurysms develop along the straight portion of these arteries. In one series of multiple PICA aneurysms distal to the anterior medullary segment, 20 of 27 aneurysms arose at non-branching points [19]. On the basis of embryologic development, PICA develops through the axial and longitudinal anastomosis of plexiform vascular formations and remnants of the lateral channel to supply the inferior brainstem and cerebellum [20]. Sites of anastomosis have been suggested as susceptible areas for aneurysm development along the straight portions of PICA and its branches [19]. Interestingly, no reports of anterior inferior cerebellar artery (AICA)-to-AICA communicating artery aneurysms have been described, and just one case of a ruptured AICA-to-PICA communicating aneurysm was reported during post-mortem analysis [21].

The present patient had a left PICA occluded at its origin, which reconstituted by a communication with the right PICA through two small collateral branching arteries at the telovelomedullary segment. Increased flow in the inter-PICA communicating collaterals presumably predisposed these vessels to the development of saccular aneurysms along the straight portion of these vessels at congenitally weak sites of vascular anastomosis. The intact aneurysm was clipped just above the neck to preserve flow through the inter-PICA collateral. However, due to the excellent bypass flow, the collateral vessel may be reluctant. As the telovelomedullary segment of PICA, proximal to the choroidal point, often possesses small perforating branches supplying the brainstem, revascularization was indicated; in situ side-to-side PICA-to-PICA anastomosis was performed to maintain right-to-left perfusion of the PICA-territories. The proximity, parallel course, and similar size of the adjacent PICAs greatly facilitate their anastomosis with one another. Care should be taken, however, to avoid prolonged occlusion times during this anastomosis as small PICA perforating vessels (< 1 mm) may be at risk. However, a longer clamping time has not been shown to have negative impact on clinical outcome in larger studies [22].

Conclusions

This unique case of inter-PICA communicating collateral aneurysm is, to our knowledge, the only reported case where multiple aneurysms are present on separate adjacent collateral arteries, with one PICA being occluded proximally and reconstituting secondary to the collateral arteries, and where, beyond aneurysm surgery, in situ bypass to restore adequate flow was performed. Clinicians should be aware of the possibility of this rare variant anatomy and corresponding pathology. In cases where hypoplasia of one side of the vertebral-basilar system is present, every effort should be made to maintain perfusion to the contralateral side. In situ bypass should be considered if the collateral must be sacrificed or if its integrity is in question, and especially if the pathology is proximal to the telovelotonsillary segment of PICA, in order to avoid serious risk of brainstem ischemia due to loss of flow through proximally arising PICA perforators.

Additional Information

Disclosures

Conflicts of interest: The authors have declared that no conflicts of interest exist.

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∗M.M.M. and J.M.F. contributed equally to this work. Author contributions to the study and manuscript preparation include the following. Conception and design: Mortazavi, Sekhar. Acquisition of data: Frerich, Mortazavi. Analysis and interpretation of data: Frerich and Mortazavi. Drafting the article: Frerich, Mortazavi, and Sekhar.
References