Technical Note

Extracranial aneurysms of the distal posterior inferior cerebellar artery: Resection and primary reanastomosis as the preferred management approach

Markus Chwajol, Ziad A. Hage, Sepideh Amin-Hanjani, Fady T. Charbel

Department of Neurosurgery, University of Illinois at Chicago, Chicago, Illinois, 60612, USA

E-mail: Markus Chwajol - markusneuro@comcast.net; Ziad A. Hage - zhage@uic.edu; Sepideh Amin-Hanjani - hanjani@uic.edu; *Fady T. Charbel - fcharbel@uic.edu

*Corresponding author

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Abstract

Background: Extracranial aneurysms of the posterior inferior cerebellar artery (PICA) are rare, with only 22 reported cases in the English literature. For saccular extracranial distal PICA aneurysms not amenable to coiling, a surgically placed clip is not protected by the cranium postoperatively, and can be subject to movement in the mobile cervical region. Furthermore, fusiform or complex aneurysms cannot be clipped primarily. Resection and primary reanastomosis is a useful surgical approach not previously described for these extracranial lesions.

Case Description: We report three cases of extracranially located distal PICA aneurysms successfully treated with this surgical strategy at our center. One patient harboring a broad necked saccular aneurysm originally underwent successful primary clipping of the aneurysm but sustained a second subarachnoid hemorrhage (SAH) on postoperative day 25 due to clip dislodgement from vigorous neck movement. The other two patients were found to have fusiform and complex aneurysms, respectively. All three patients were ultimately treated with resection and end-to-end PICA anastomosis, which successfully obliterated their aneurysms.

Conclusions: Resection and primary reanastomosis of extracranial distal PICA aneurysms averts the risk of clip dislodgement due to neck movement and/or compression by soft tissues in the upper cervical region. It is a safe and efficacious technique, which we propose as the preferred management strategy for these rare vascular lesions.

Key Words: Anastomosis, aneurysm, PICA, resection, subarachnoid hemorrhage

INTRODUCTION

Posterior circulation aneurysms make up for 5-9% of all intracranial aneurysms. The majority of these lesions originate at or around the basilar tip complex and only about 1% of all intracranial aneurysms are posterior inferior cerebellar artery (PICA) lesions, of which most are located at the vertebral artery (VA)–PICA junction or proximal PICA. Distal PICA aneurysms are even less common, and can occur extracranially. Overall, only 22 lesions reported in the literature have been located extracranially, and of these, only 11 were distal aneurysms.
Of all the reported 11 distal extracranial lesions, one was treated with endovascular coiling; all other lesions were treated with surgical clip obliteration. However, an aneurysm clip is not protected by the cranium after suboccipital craniectomy (SOC) and C1 laminectomy are performed as part of the surgical procedure. Therefore, postoperatively patients may be at risk for exposing the clip to shifts or pressures from overlying soft tissues and/or head movement.

In this article, we illustrate a case in which head motion during rehabilitation was implicated in clip dislodgement after surgical clipping of a ruptured distal extradural PICA aneurysm, with resultant rebleeding. We discuss resection and primary reanastomosis as a valuable surgical approach to avoid this complication, as well as to treat fusiform and complex distal extracranial PICA aneurysms. We review the clinical indications, radiographic characteristics, and surgical outcomes in our series of three patients. In addition, we also provide a thorough review of the literature on distal extracranial PICA aneurysms and their surgical treatment, including the significance of the origin and course of the parent vessel.

**CASE REPORTS**

Review of all aneurysm cases performed between 2001 and 2009 revealed that three cases of distal extradural ruptured PICA aneurysms were treated by the senior author at our institution during this time period. The clinical indications and radiographic characteristics are presented below.

**History and examination**

**Case 1**

A 76-year-old Hispanic female suddenly collapsed and was brought to the emergency room of an outside hospital where a head computed tomography (CT) revealed diffuse subarachnoid hemorrhage (SAH) with blood in the fourth, third and lateral ventricles. Upon arrival at our institution, she was a Hunt and Hess grade III SAH. Repeated CT showed the SAH and hydrocephalus [Figure 1a and b] and right frontal external ventriculostomy drain (EVD) was placed emergently. A diagnostic cerebral angiogram demonstrated the presence of a right PICA aneurysm measuring approximately 2-2.5 mm in diameter with a broad neck [Figure 1c]. Decision was made that due to its morphology, the aneurysm should be treated with microsurgical clipping.

A SOC with C1 and partial C2 laminectomies was performed. The cisterna magna was found to be full of blood; it was explored and the hematoma evacuated. Both segments of the PICA, proximal and distal to the aneurysm, were identified. The caudal loop of the PICA was seen under the C2 lamina and the aneurysm itself

![Figure 1a: CT scan of the head in axial cuts showing diffuse subarachnoid hemorrhage, intraventricular hemorrhage and hydrocephalus](image1.png)

![Figure 1b: CT scan of the head in axial cuts showing diffuse subarachnoid hemorrhage, intraventricular hemorrhage and hydrocephalus](image2.png)

![Figure 1c: Digital subtraction angiogram (DSA) in AP projection demonstrating the presence of a right PICA aneurysm measuring approximately 2-2.5 mm in diameter with a broad neck (black arrow)](image3.png)
projected under the lamina of C1, and was located on the tonsillomedullary segment. Under temporary clipping distal and proximal, the aneurysm neck was defined and the aneurysm was clipped. The dura was then closed at the more distal cervical region, and Duragen (Integra LifeSciences Corporation, NJ) was placed more cranially. Cervical fascia and skin were closed in the usual fashion.

The patient recovered well from the surgery and passed EVD challenge. CT on postoperative day (POD) 19 revealed resolution of the SAH, and no blood in the posterior fossa around the aneurysm clip. Postoperative skull X-ray (XR) showed the clip located below the foramen magnum [Figure 1d]. The patient was scheduled to be discharged to an inpatient rehabilitation facility, and on POD 25, following a physical therapy session including active range of motion of bilateral upper extremities, was found unresponsive on the floor. Emergent CT scan of the head showed the presence of blood in the fourth, third and lateral ventricles, and right frontal EVD was placed. A repeat diagnostic angiogram revealed findings consistent with rebleed from the previously clipped right PICA aneurysm.

Case 2
This is a 70-year-old female who was found unresponsive, and was brought to an outside hospital where head CT showed SAH in the basal, peripontine, and foramen magnum cisterns, as well as intraventricular hemorrhage (IVH) in the occipital horns, third and fourth ventricles. The patient arrived as Hunt and Hess grade III SAH. A repeat CT demonstrated moderate enlargement of the ventricular system correlating with hydrocephalus and a right frontal EVD was placed. A four vessel cerebral angiogram showed an approximately 9 × 9 mm left PICA aneurysm located at the level of the caudal loop (tonsillomedullary segment). The aneurysm was single lobed, fist shape in appearance, angiographically located at the skull base, likely at the level of the VA dural penetration, with what appeared to be both intracranial and extracranial components [Figure 2a and b]. Subselective angiography demonstrated separate inflow and outflow branch vessels [Figure 2c]. Coil embolization was attempted but the procedure was aborted as it was felt that there was high risk of a left PICA occlusion and subsequent ischemic infarct.

Case 3
The patient is a 52-year-old right handed African-American woman who suffered a sudden onset of severe headache and neck pain with multiple episodes of vomiting. Head CT at an outside hospital showed mild amount of SAH in Sylvian fissures and temporoparietal cortical sulci bilaterally, and intraventricular blood within the fourth, and left lateral ventricles, and mild hydrocephalus. The patient had a magnetic resonance angiogram (MRA) done the following day, which was unremarkable for any vascular abnormality, however, a conventional cerebral
Figures 2a: DSA in AP (2a) and lateral (2b) projections showing an approximately 9 × 9 mm left PICA aneurysm (black arrow) located at the level of the caudal loop (tonsillomedullary segment). The aneurysm appears to have both intracranial (1) and extracranial (2) components in Figure 2b.

Figure 2c: Subselective angiography of the left PICA aneurysm in oblique projection demonstrating separate inflow (black arrow) and outflow (white arrow) branch vessels.

Figure 2d: Intraoperative photo showing the aneurysm surrounded by clot (black arrow) and the left PICA inlet (1) and outlet (2) segments. A perforator coming off of the inlet segment is visualized as well (3).

Figure 2e: Intraoperative photo showing the completed end-to-end PICA anastomosis (black arrow) after the aneurysm was excised.

Figures 2f: Postoperative DSA in AP (2f) and lateral (2g) projections showing no aneurysmal remnant and a patent left PICA. The white arrow shows the end-to-end PICA anastomosis site.
angiogram done subsequently was suspicious for a left PICA aneurysm. She was transferred to our institution for further management. Upon admission to our center, she was a Hunt and Hess grade II SAH with only a very mild left-sided arm and leg weakness. A four vessel cerebral angiogram was performed, which showed an $8 \times 6$ mm fusiform left PICA aneurysm at the tonsillomedullary segment (caudal loop) behind the arch of C1, not amenable to endovascular coiling [Figure 3a-c].

**Operation and postoperative course**

In all three cases, the patients underwent SOCs and C1/C2 laminectomies with resection of the aneurysms and primary reanastomosis. The details of the surgical approach and outcomes are presented below.

**Case 1**

The patient was taken to the operating room for reexploration of the previously clipped right PICA aneurysm. In the operating room, the cervical fascia was reopened and the previous SOC was identified. A moderate amount of blood clot was present deep to the muscle and this was evacuated. The aneurysm clip was identified and found to be protruding through the layer of Duragen (Integra LifeSciences Corporation, NJ) previously used to cover the remaining dural defect [Figure 1e]. The hematoma was evacuated from around the Duragen (Integra LifeSciences Corporation, NJ), which was then removed, and the clipped aneurysm was exposed. During the exploration, it appeared that the clip had become dislodged from its original placement at the neck of the aneurysm. Intraoperative bleeding was encountered, and controlled by placement of temporary clips on the PICA proximally and distally to the aneurysm. The aneurysm was resected with the use of microdissecting scissors. Both arterial ends were then spatulated and reanastomosed with running 9-0 nylon on the posterior wall and interrupted 9-0 nylons.
on the anterior wall. After removing the temporary clips, adequate antegrade flow in the distal PICA was confirmed with the transonic flow probe (Transonic, NY), and found to be approximately 35-40 ml/min. Duragen (Integra LifeSciences Corporation, NJ) was again placed to cover the dural defect and the fascia and skin were closed as previously. Postoperative angiogram demonstrated a patent anastomosis and good filling of the distal PICA.

Postoperatively, the patient did well, and later underwent a right frontal ventriculo-peritoneal shunt placement. CT 14 days after the reoperation showed full resolution of the SAH. Twenty-eight days after her second surgery the patient was found unresponsive on the floor with bilateral blown pupils. Emergent CT scan showed an unrelated massive left thalamic bleed with IVH. Due to extremely poor prognosis the family decided to withdraw further care and disconnect the patient from the ventilator. The patient expired 6 days later.

Case 2
The patient underwent a left SOC with C1 laminectomy. Immediately upon opening the dura, a large area of thick acute clot was seen in the area of the foramen magnum and it was carefully suctioned out. Following visualization of the aneurysm, the left PICA was noted to be going into and out of the aneurysm, thus forming a separate inlet and outlet. Irregular, grossly abnormal arterial vasculature in between the inlet and outlet was identified. In addition, significant perforators coming off of the inlet and outlet of the left PICA were noted, making vessel sacrifice high risk. Based on these findings, it was decided that excising the aneurysm and performing a left PICA primary reanastomosis would be the best strategy.

Temporary clips were placed proximal and distal to the aneurysm. Microscissors were used to excise the aneurysm en block at the distal parts of the inlet and outlet. After this, the edges of the inlet and outlet vessels were trimmed and fish-mouthed, and an end-to-end PICA anastomosis was performed as described in the first case. Good filling of the distal PICA was noticed after removal of the temporary clips. Flow just distal to the reanastomosis was measured with the intraoperative flow probe and was 18.5 cc/min, which was consistent with initial flow measurements. A postaneurysmectomy indocyanine green (ICG) angiography demonstrated good flow in the bypass as well as evidence of flow in the perforators. A postoperative angiogram performed the same day showed no aneurysmal remnant and a patent left PICA.

The patient then had a right ventriculo-peritoneal shunt placed on POD 8, and was discharged to acute rehabilitation facility on POD 15, with only mild residual left lower extremity weakness and minimal residual memory loss. She eventually recovered to her prehospitalization baseline.

Case 3
A SOC and C1, and partial superior C2 laminectomies were performed. After opening of the dura, the arachnoid was dissected under a microscope and the fusiform left PICA aneurysm with its PICA inlet and outlet was localized at the midtonsillar level. Intraoperative flow probe measurements revealed a flow of 25 cc/min in the parent vessel. Temporary clips were placed, proximal and distal to the aneurysm. The fusiform aneurysm was then resected with microscissors. The fish mouthing technique was again used on each side of the vessel and reanastomosis of both ends using 8-0 nylon sutures was performed. Postanastomosis flow measurements demonstrated a well preserved flow of 19 cc/min. ICG video angiography demonstrated left PICA midtonsillar segment without the aneurysm and a patent anastomosis.

A postoperative angiogram showed robust filling of the anastomosis and no evidence of aneurysmal remnant. Table 1 includes all 11 distal extracranial PICA aneurysms previously reported in the English literature in addition to 3 cases from the present report. The average patient age in previously reported 11 cases was 36.4 years, the average age of our cohort was 66 years, and the combined average age of all 14 patients was 42.7 years, respectively. IVH in the fourth ventricle was found in all ruptured PICA aneurysms when CT
data was available (N = 12). Female to male ratio was 1.3. Hunt and Hess grade distribution was as follows: 0 – 0 patients, I – 2 patients, II – 4 patients, III – 7 patients, IV – 1 patient; V – 0 patients. PICA origin was intradural in eight cases (57%), extradural in two cases (14.5%), and unspecified in four cases (28.5%). EVD placement was required in five cases (36%), not required in six cases (43%), and was not specified in three cases (21%). Three cases (21%) resulted in permanent ventriculoperitoneal shunt (VPS) placement, six cases (42%) did not necessitate VPS, and it was unspecified in five cases (37%). The distribution of clinical outcomes was as follows: excellent/return to baseline (neuro-intact) – 10 cases (71%), minor deficit/living independently – 2 cases (14.5%), and mortality – 2 cases (14.5%).
PICA aneurysms represent 0.5-3% of all cerebral aneurysms.\textsuperscript{20,30,31,41} They usually originate at the VA–PICA junction.\textsuperscript{7,25,30,31,36} and only about 15-30% are peripheral PICA aneurysms.\textsuperscript{20,22} Cases of PICA aneurysms are infrequently reported and reviews are also rare.\textsuperscript{2,7,39} Only 22 extracranial PICA aneurysms have been reported in the literature. In a review of 228 PICA aneurysms, 3 were found extracranially\textsuperscript{2} and in another review,\textsuperscript{30} of 26 lesions, only 1 extracranial aneurysm was found. The rest were single aneurysm case reports.\textsuperscript{1,3,5,7,11,16-19,23,28,37,40,42-45}

**PICA anatomy**

There are significant variations in PICA origin and course.\textsuperscript{1,11,29,36,41} Extracranial origin of PICA was found in 18% cadavers,\textsuperscript{29} and 5-20% of PICA take offs are extradural.\textsuperscript{1,14,38} One cadaveric study showed the origin to be as low as 14 mm below the foramen magnum in 7 of 42 PICAs studied in 25 adult cadavers.\textsuperscript{29} When the origin is extracranial, it is usually from the V3 segment of VA\textsuperscript{14} and 6-17.9 mm below the foramen magnum.\textsuperscript{1,28} When the origin is posterior to the posterior arch of C1, it is likely to be extradural.\textsuperscript{14} and in such cases, the PICA course stays lateral and posterior. In the case of extracranial PICA origin, the caudal loop is located below the foramen magnum or absent, and typically no branches to brainstem are present from the extracranial segment of PICA.\textsuperscript{1,14,44}

Extradural PICA origin is usually not associated with PICA aneurysms but both intra- and extradural aneurysms have been reported \[Table 1\]. The variability and complexity of PICA anatomy makes aneurysms appear in unusual locations, and there is great variation in shape and size of the lesions.\textsuperscript{10} Extracranial PICA aneurysms are frequently associated with aberrant PICA anatomy; lack of caudal or/and cranial loops is frequently observed, and PICA may cross to the contralateral site.\textsuperscript{28,44} \[Table 1\]. It seems that hemodynamic stress plays an important role in the formation of distal PICA aneurysms,\textsuperscript{15} including those formed outside of the cranium. Out of the 14 distal extracranial PICA aneurysms (including our series), 13 (93%) were located at the caudal loop/tonsillomedullary segment. This suggests that the hairpin turn of the caudal loop may act as a branching point with increased hemodynamic stress and increased probability of aneurysm formation at this location.

**Treatment options**

Coil embolization of intracranial proximal PICA aneurysms is a safe and efficacious treatment strategy.\textsuperscript{31} Intracranial distal PICA lesions have also been successfully treated with endovascular approaches.\textsuperscript{47} In contrast, all reported cases of distal extracranial PICA aneurysms have indicated surgical treatment. Endovascular access and therapy might be difficult due to the aberrant origin and course of the parent vessel, its tortuosity, as well as the aneurysm shape that frequently is not suitable for coil embolization.\textsuperscript{34,46} Surgery for clipping of extracranial PICA aneurysms can be challenging due to similar considerations: complexity and variability of PICA and VA anatomy, unusual shapes and rarity of the aneurysms, and their close anatomical relationship to the foramen magnum, and C1 arch, although access to the lesion and proximal control is typically more direct in such cases.

Detailed knowledge of the vascular anatomy before surgery is imperative. Serious consideration should be given to performing a preoperative angiogram with subselective angiography. Additional information gained from these procedures might help delineate the vascular anatomy better, and reveal the anatomical relationships of the vascular and bony structures in the region. It is particularly important to identify the location of the aneurysm in relation to the foramen magnum and arches of C1 and C2 vertebral bodies. Computed tomography angiogram (CTA) with 3D reconstruction is an excellent modality to elucidate the regional anatomy of both the vessels and bones during preoperative planning. Both VA and PICA extracranial anatomy needs to be carefully reviewed before surgery.\textsuperscript{5} It is critical for the surgeon to realize that when an aneurysm is located below or in the close proximity to the C1 arch, the probability of intraoperative rupture during C1 laminectomy might be high.

Familiarity with important surgical concepts is also crucial for successful outcome. If the PICA origin is extradural, temporary clip placement on the VA after dural opening would not serve its intended purpose. External ventriculostomy or lumbar drains are usually not required to divert the cerebrospinal fluid, as cisterna magna can easily be opened and good brain relaxation be achieved during surgery.\textsuperscript{57}

Intracranial and extracranial fusiform, atherosclerotic, and giant aneurysms of PICA are surgically challenging lesions. The notion of vessel reconstruction and anastomosis as well as bypass for flow augmentation and/or flow replacement has been well established for over two decades now.\textsuperscript{4,6,8,67} Surgical management with aneurysm trapping and end-to-side PICA–PICA anastomosis for the treatment of a VA-proximal PICA aneurysm has been previously discussed.\textsuperscript{47} Moreover, the concept of resection and end-to-end reanastomosis has been reported for the treatment of intracranial giant PICA aneurysms located at the caudal loop.\textsuperscript{15,32} In addition, two giant aneurysms of the caudal PICA loop were excised without reanastomosing the parent vessel.\textsuperscript{35,48} Although sacrifice of the PICA beyond the caudal loop may be reasonably well tolerated due to relative lack of brainstem perforators in this region, the extent of cerebellar stroke is not well predictable, and vessel preservation is preferable.

Applying the same concept of resection and end-to-end reanastomosis to broad neck and fusiform distal
extracranial PICA aneurysms has not been reported. Here we present three cases of caudal PICA loop aneurysms treated with this surgical approach, to highlight the particular suitability and advantages of this approach for this location as a preferred option, even if the aneurysm is deemed amenable to primary clipping. The advantage is best illustrated by the first patient who initially underwent successful primary clipping. She sustained a second SAH during her hospitalization after a physical therapy session, suspected to be due to clip dislodgement by the overlying soft tissues during neck movement. This hypothesis was confirmed during the second surgery, as fresh blood clot was found around the clip and aneurysm neck, and the clip was found protruding through the dural substitute. The aneurysm was resected and end-to-end anastomosis was successfully performed, with good initial postoperative outcome.

The two other patients further demonstrate the feasibility of this approach, and its ease of application due to the superficial and easily accessed nature of these distal PICA extracranial aneurysms. Furthermore, the location (distal PICA) of these aneurysms, being a relatively perforator-free zone, renders this approach even more attractive. These two patients were found to have fusiform and complex PICA aneurysms, respectively, with neither of the aneurysms being amenable to coiling. In both cases, we performed resection of the aneurysms with primary reanastomosis as described in the first case. Both patients did well postoperatively and returned to their preoperative baselines.

We used the midline SOC approach with C1 laminectomy (second case), and C1–C2 laminectomies in the first and third cases. In all three cases, we performed careful preoperative analysis of the bony anatomy as it related to the aneurysm and the parent vessel, and avoided intraoperative rupture during bone removal. We believe that analysis of unsubtracted angiographic images is adequate for this purpose in most instances; nonetheless, CTA remains a great modality choice should any uncertainty exist.

In conclusion, resection and primary reanastomosis of distal extracranial PICA aneurysms is a safe and efficacious surgical approach. We propose this as the treatment of choice for these rare vascular lesions when surgical management is needed, compared with direct clipping, as this strategy avoids the risk of clip repositioning or dislodgement due to neck movement and/or compression by soft tissues outside of the cranium.

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