

Remote distal outflow occlusion: a novel treatment option for complex dissecting aneurysms of the posterior inferior cerebellar artery

Report of 3 cases

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The authors report a novel management option for patients with complex dissecting aneurysms of the posterior inferior cerebellar artery (PICA).

The authors reviewed the medical records and neuroimaging studies of 3 patients who underwent a novel surgical treatment for complicated dissecting PICA aneurysms. The mean follow-up period was 1.1 years, and no patient was lost to follow-up. Two patients were in poor condition following an acute, severe subarachnoid hemorrhage, and 1 presented with headaches and a remote history of bleeding. All patients underwent surgical occlusion of the PICA beyond the tonsillar loop, distal to the aneurysmal segment. Intraoperative and delayed follow-up angiography demonstrated progressive diminution in size of the aneurysmal dilation but persistent filling of the proximal PICA segments supplying the brainstem. Outcome was good in all cases.

This novel technique has been used successfully in 3 cases and, to the authors' knowledge, has not been reported previously. (DOI: 10.3171/2009.1.JNS081250)

KEY WORDS • aneurysm • brain • bypass • dissection • posterior inferior cerebellar artery • stroke

DISSECTING aneurysms of the PICA are extremely rare lesions that have been described in only limited fashion in the neurosurgical literature.^{1–3,5,6,10,11,14,17,18,20–24} We have previously described a series of 6 patients with dissecting PICA aneurysms and included long-term clinical and angiographic follow-up data.¹⁸ These patients were managed with either circumferential wrap/clip reconstruction or proximal occlusion combined with distal revascularization. Currently, these 2 techniques remain our preferred treatment options for such aneurysms. Nevertheless, we have now encountered 3 patients for whom these options did not seem feasible, and a novel, alternative surgical strategy, which we have termed “remote distal outflow occlusion,” was used. We describe the technique, results, and the rationale behind the technique's use.

Abbreviations used in this paper: PICA = posterior inferior cerebellar artery; VA = vertebral artery.

Case Reports

During the 18-month period between January 2007 and June 2008, our neurovascular service treated 496 intracranial aneurysms. Of these, 5 were dissecting aneurysms involving the PICA. We retrospectively reviewed the records of 3 patients with complicated PICA dissecting aneurysms managed by remote distal outflow occlusion. Hospital records, neuroimaging studies, operative reports, and follow-up clinic notes were available in all cases. The patients were followed for 5–18 months (mean follow-up period 1.1 years).

All patients underwent surgical exploration of the aneurysms via a far-lateral suboccipital craniotomy in which C-1 laminectomy and partial condylar resection were performed. The ipsilateral VA was identified extradurally and then traced intradurally to the PICA origin. When proximal control was achieved, the dissected segment was then explored. Mild hypothermia (34°C) and



FIG. 1. Case 1. A and B: Lateral VA arteriographic and 3D rotational images showing the very irregular dissecting PICA aneurysm (arrow) with dramatic contrast extravasation providing an outline of the spinal canal (arrowheads). The distal VA is atretic beyond the PICA (asterisk). C and D: Follow-up arteriograms obtained 1 day after remote distal outflow occlusion demonstrating persistent filling of the proximal PICA segments (arrows) with dramatic, complete resolution of the aneurysm. E: Axial diffusion weighted MR image revealing limited ischemic injury in the distal PICA distribution.

barbiturate anesthesia were used in all cases. In all 3 patients, angiography demonstrated the dissecting aneurysms to involve the proximal PICA beginning just beyond its origin from the VA. There was significant irregularity of the vessel, which normalized before the tonsillar point in every case. The details of surgery follow. As part of the closure, a generous duraplasty was performed.

Case 1. This 43-year-old man developed severe headache followed by transient loss of consciousness. At an outside emergency department, he was awake and talking but suddenly developed severe hypertension, loss of consciousness, and decerebrate posturing movements. He was intubated and transferred to our center. Emergency cerebral angiography demonstrated an irregular dissecting aneurysm of the right PICA with active contrast extravasation filling the cervical canal with dye (Fig. 1A and B). Recombinant activated Factor VIIa was administered, and the patient was taken directly to the operating room where he underwent emergency ventricular drainage followed by far-lateral suboccipital craniotomy. A thick clot was evacuated from the cisterna magna, and the right PICA was identified at the tonsillar loop. The involved vessel was bluish well beyond the aneurysm. The contralateral PICA was atretic, and no attempt had been made to preserve the occipital artery because of the urgent nature of the procedure, which was performed in the middle of the night.

Rather than attempting to manipulate the dissected segment to undertake wrapping, a decision was made to occlude the PICA just distal to the tonsillar loop. Postoperative angiography 8 hours later demonstrated opacification of the proximal PICA segments and no filling of the aneurysmal dilation (Fig. 1C). Postoperative MR imaging showed limited evidence of diffusion changes in the distal PICA territory and no evidence of brainstem ischemia (Fig. 1D). The patient was maintained in barbiturate-induced coma for 72 hours. He subsequently awoke without focal deficit. He required revision of his duraplasty for a postoperative CSF leak and placement of a ventriculoperitoneal shunt for delayed hydrocephalus, but otherwise he made an excellent recovery.

Case 2. This 47-year-old woman was found to have a large aneurysm associated with a PICA dissection that had bled 6 weeks prior to referral to our center (Figs. 2A and B). At the time of surgical exploration, the aneurysm was densely adherent to the lower cranial nerves and brainstem, precluding safe circumferential wrapping (Fig. 2C). There was significant size asymmetry between the right and left distal PICAs, which would have made side-to-side anastomosis difficult, and the occipital artery had been injured during the opening.

As a result, we chose to perform remote distal outflow occlusion of the PICA beyond the tonsillar loop, occlud-

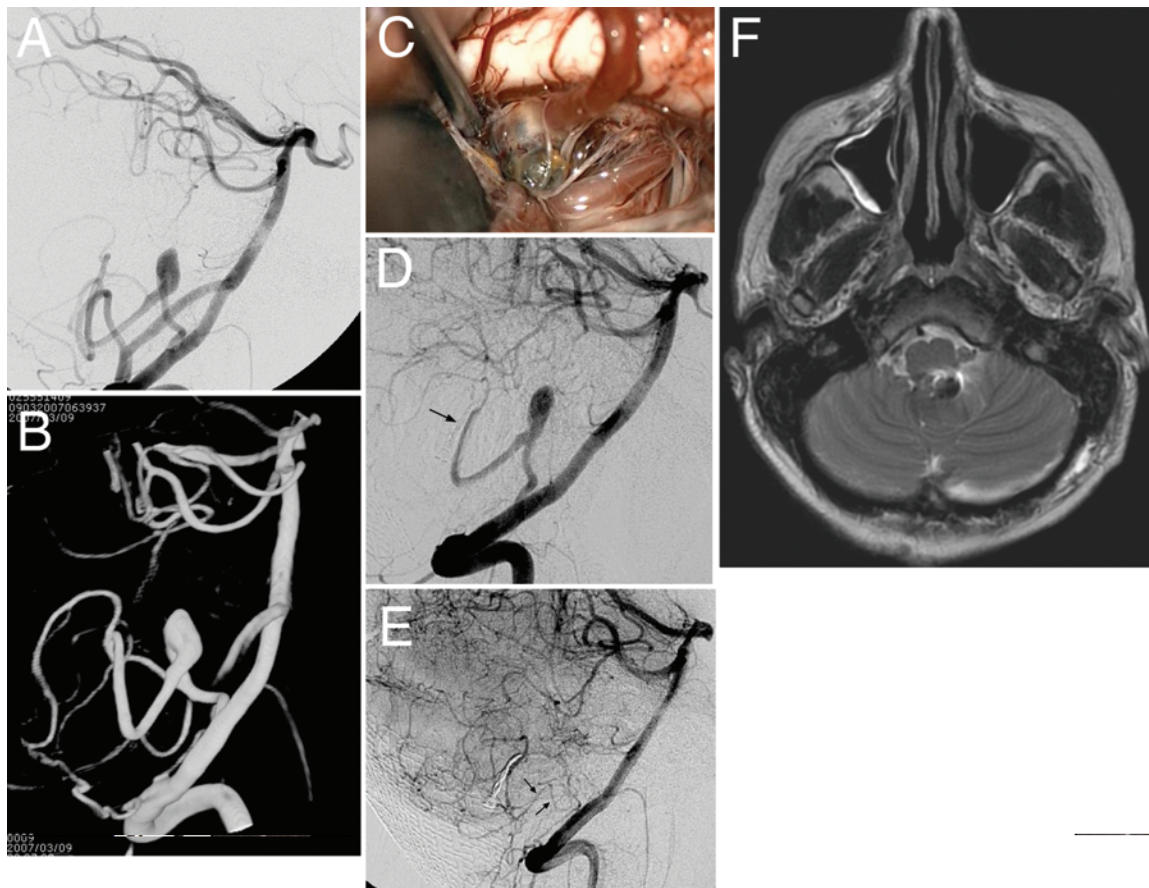


FIG. 2. Case 2. A and B: Lateral and 3D rotational images of a VA arteriogram demonstrating a large irregular aneurysm involving the PICA. C: Intraoperative photograph showing the irregular dissecting aneurysm, which is partly thrombosed and intimately adherent to the lower cranial nerve fibers, precluding safe circumferential wrapping. D: Intraoperative arteriogram revealing clip occlusion of the distal PICA (arrow) with sparing of a large cerebellar branch. E: Follow-up arteriogram obtained 1 month later showing dramatic resolution of the aneurysm with persistent filling of the PICA itself (arrows). F: Axial T2-weighted MR image revealing no evidence of PICA distribution ischemia.

ing the PICA just distal to a large cerebellar branch. Intraoperative angiography demonstrated slight diminution in the size of the aneurysm as well as preserved filling of the proximal PICA and large cerebellar branch (Fig. 2D). The patient awoke without neurological deficit, and postoperative MR imaging demonstrated no diffusion changes.

Follow-up angiography performed 1 week later demonstrated the aneurysm to be smaller than it had been preoperatively but slightly larger than on the intraoperative study. Subsequent angiography at 1 month indicated that the aneurysm was completely thrombosed with filling of a very small residual PICA (Fig. 2D). Follow-up MR imaging remained unremarkable (Fig. 2E). With the exception of persistent headaches, the patient had no other problems.

Case 3. This 34-year-old woman presented with acute severe subarachnoid hemorrhage, hemorrhage within the fourth ventricle, and associated hydrocephalus. Her neurological status was Grade IV Hunt and Hess preoperatively, with bilateral pupillary dilation, and she underwent surgical treatment on an urgent basis after bleeding twice prior to arrival at our center and again during diagnostic angiography, the images of which suggested the presence of a dissecting aneurysm of the PICA (Figs. 3A and B). At the time

of surgery, significant swelling of the cerebellum was encountered. As a result, we believed distal revascularization would be quite difficult. In addition, we chose not to wrap the aneurysm because we did not wish to directly manipulate the dissected segment, which had bled repeatedly.

We removed the ipsilateral cerebellar tonsil using subpial dissection to properly expose the involved PICA. The PICA itself was bluish-black and diffusely dilated until it became more normal in appearance just proximal to the tonsillar point. A decision was made to surgically occlude the PICA immediately distal to the last visible brainstem perforating artery, which arose approximately 3 mm beyond the tonsillar point. Intraoperative and delayed postoperative angiography confirmed occlusion of the PICA with minimal slow filling of the proximal PICA segments and limited opacification of the aneurysmal dilation (Fig. 3C). The patient was maintained in barbiturate-induced coma for 4 days, and then she gradually awoke. Postoperative MR imaging demonstrated no evidence of brainstem ischemia with limited diffusion-weighted changes in the distal PICA territory (Fig. 3D).

The patient required a tracheostomy and placement of a ventriculoperitoneal shunt, followed by prolonged inpatient rehabilitation, but she made a striking recovery

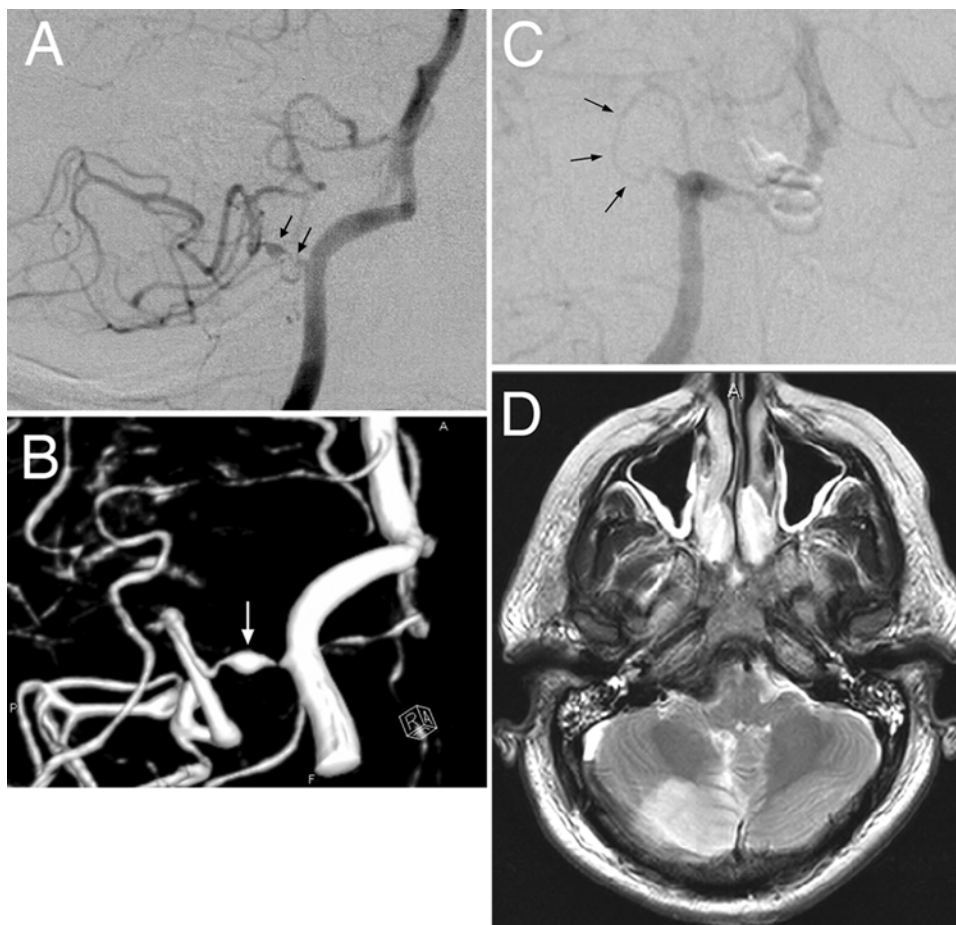


FIG. 3. Case 3. A and B: Preoperative lateral and 3D rotational angiographic images demonstrating an irregular fusiform abnormality (arrows) of the proximal PICA compatible with a dissecting aneurysm. C: Postoperative VA arteriogram obtained 1 week after surgery showing limited residual filling of the proximal PICA following distal clip occlusion. D: Axial T2-weighted MR image revealing a small peripheral PICA distribution infarct.

and was able to regain full functional independence. Delayed cerebral angiography demonstrated persistent filling of an atretic remnant of the proximal PICA without opacification of the aneurysmal dilation.

Discussion

Dissecting aneurysms of the PICA are extremely rare lesions that tend to be highly unstable and carry a significant risk of repeated hemorrhage.^{1,6,7,18,22} The optimal management of dissecting PICA aneurysms remains controversial. Although, the majority of cases described in the literature have been treated by proximal PICA occlusion or trapping of the involved segment, more recent reports have increasingly described endovascular PICA occlusion as an alternative option.^{1,14,21,22,24} Because these techniques carry the potential risk of devastating brainstem ischemic injury, we have previously treated these lesions with either circumferential wrap/clip reconstruction, preserving the parent artery when possible, or with distal revascularization when proximal occlusion was deemed necessary.¹⁸ In 3 cases, however, we were unable to achieve either of these options, and we used a novel approach, which forms the basis for this report.

Rationale for Remote, Distal Outflow Occlusion

The idea that an intradural aneurysm could be safely treated by distal occlusion was first suggested to the senior author (E.S.N.) by Dr. Robert Spetzler during a conversation regarding the treatment of a complex posterior circulation aneurysm (personal communication, 2001). Although the idea of occluding the efferent outflow of an aneurysm, particularly following rupture seemed counterintuitive initially, further reflection suggested the possible merits of such an approach. Cardiothoracic, vascular, and neurovascular surgeons who perform bypass procedures with regularity are intimately familiar with the devastating consequences to an anastomosis of “poor distal runoff.” Outflow occlusion produces an immediate diminution in the amount of flow through the occluded artery, and although the pressure within the aneurysm may momentarily increase, it should rapidly diminish thereafter.

Isolated case reports have described the use of distal occlusion in the management of partially thrombosed, giant, fusiform M₁ segment aneurysms.^{4,8,12} In these reports, proximal occlusion or trapping of the lesion had been planned preoperatively, but at the time of surgical exploration, proximal occlusion was found to be unacceptably dangerous or was left as a possible future option,

and extracranial-intracranial bypass with distal occlusion alone was followed by rapid aneurysm thrombosis.^{4,8,12} Based on the Bernoulli equation, Horowitz et al.⁸ created an elegant mathematical model describing the intraluminal pressure changes that might be expected following distal aneurysm occlusion. They demonstrated that the resulting variations in pressure should be less than those induced by normal daily activities and concluded that distal occlusion would not be expected to increase the risk of aneurysm rupture.^{4,8} Nevertheless, we suspect that a general discomfort with the idea of suddenly occluding the outflow of an aneurysm has limited both the actual use and an awareness of this technique.

The challenges associated with the management of dissecting aneurysms of the PICA have been discussed previously.¹⁸ The dissected segment itself typically gives rise to brainstem perforating vessels. As a result, proximal occlusion or trapping procedures risk ischemic injury to the brainstem. In the past, we have treated these aneurysms with either circumferential wrap/clip reconstruction or proximal occlusion with distal revascularization using side-to-side PICA-PICA anastomosis or occipital artery-PICA bypass.¹⁸ Nevertheless, as detailed above, we have now encountered 3 cases of complex dissecting PICA aneurysms in which these options seemed dangerous or impractical.

In these cases, we hypothesized that remote, distal outflow occlusion, sacrificing the PICA beyond the tonsillar loop, might be an excellent treatment option. In all cases, the PICA was occluded well beyond the aneurysmal dissection and beyond the last visible brainstem perforating vessel. In theory, it was believed that a limited amount of flow should continue through the proximal PICA segments to irrigate the small brainstem perforating branches arising from these segments. At the same time, it was thought that this small amount of flow would not be enough to risk further bleeding or expansion of the dissecting aneurysm. In all cases, the distal occlusion appeared to substantially diminish flow through the aneurysmal segment resulting in diminution in size followed by complete resolution of the aneurysm. Delayed angiography demonstrated limited persistent filling of the proximal PICA segments, which appeared to successfully avoid brainstem ischemic injury in all cases.

Because these dissecting aneurysms almost uniformly involve the proximal PICA segments and incorporate brainstem perforators into the aneurysmal segment itself, simple proximal occlusion may carry an unacceptable risk of severe brainstem injury. As has been noted in the past, PICA occlusion distal to the telovelotonsillar segment generally does not result in brainstem injury and is well tolerated by most patients.^{1,9,11,13,15,16,22} Therefore, distal occlusion, sacrificing the PICA beyond the last brainstem perforator, should generally limit the risk of ischemic injury to the distal cerebellar territory supplied by the PICA. A generous duraplasty was performed as part of the procedure in all 3 cases to provide decompression in the event a significant PICA infarct developed. In fact, none of our patients had significant cerebellar edema due to a large ischemic injury.

When practical, we have argued for attempted dis-

tal revascularization in cases of PICA sacrifice because of the uncertain adequacy of collateral supply both to protect the brainstem and also to avoid infarction of the cerebellum.^{18,19} Although distal PICA infarction is not disabling in most cases, postoperative cerebellar edema and cerebellar dysfunction are unnecessary if revascularization can be performed successfully. Some patients have a very large PICA that supplies the anterior inferior cerebellar artery territory, as well. For those patients, PICA sacrifice, even distal to the tonsillar point, may be dangerous because of the severe cerebellar edema that may develop. Given this, we present distal occlusion as one of several options in the management of this difficult disease process. If possible, we still favor circumferential wrap/clip reconstruction or proximal occlusion with distal revascularization as primary options in this setting. Nevertheless, we now consider remote distal outflow occlusion to be an important alternative when these options appear impractical.

Limitations of the Present Study

It should be noted that remote distal outflow occlusion does not definitively exclude the involved segment from the arterial circulation and thus cannot completely eliminate the potential for repeated hemorrhage.³ Nevertheless, serial angiographic evaluations of all the aneurysms treated in our series demonstrated their rapid diminution in size followed by complete obliteration. We suggest that the limited residual filling of the dissected segment should provide little impetus for delayed enlargement of the aneurysmal dilation or for rebleeding. In some cases, distal occlusion could result in retrograde thrombosis of the vessel, potentially placing brainstem perforators at risk, although we did not encounter this problem in our 3 cases. To our knowledge, this type of remote distal outflow occlusion has not been reported previously. Although we have only applied this technique in 3 patients, the disease process itself is uncommon, and we therefore considered it worthwhile to describe our results.

Conclusions

We have described a novel treatment option that was used in the management of 3 patients with complex dissecting PICA aneurysms. All were treated surgically with remote, distal outflow occlusion resulting in dramatic angiographic resolution of the aneurysmal dilation. The residual filling of the proximal PICA prevented brainstem ischemic injury in all cases, and yet the limited amount of filling appears to leave little impetus for further growth or potential rebleeding from these aneurysms. The goal of the technique is to preserve perforators distal to the aneurysmal segment, and, to our knowledge, this technique of outflow occlusion remote from the aneurysm has not been described previously.

Disclaimer

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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