Ruptured traumatic vertebral artery pseudoaneurysm in a child treated with trapping and posterior inferior cerebellar artery reimplantation

Case report and review of the literature

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The authors present the case report of a pediatric patient with a ruptured traumatic pseudoaneurysm of the intracranial vertebral artery (VA) from which the posterior inferior cerebellar artery (PICA) emerged. After considering multiple therapeutic options, the patient was treated surgically by trapping of the aneurysm segment and direct reimplantation of the PICA distal to the rupture site. In addition to presenting this unique case, the authors discuss the treatment of VA pseudoaneurysms and the various techniques for PICA revascularization. A review of the literature on PICA reimplantation is provided as an adjunct in the treatment of complex VA aneurysms.

KEY WORDS • bypass • cerebral revascularization • dissecting pseudoaneurysm • posterior inferior cerebellar artery • trapping • vertebral artery • pediatric neurosurgery

PSEUDOANEURYSMS involving the intracranial VA, an increasingly recognized cause of SAH, are complex lesions with a threatening natural history.1–9,12,17,22,23,34,43,45,48,55,57,62,63 Because the vessel injury is both transmural and circumferential in nature, these lesions do not typically lend themselves to conventional clip-assisted arterial reconstruction.1–3,8,9,11,16–18,21,23,29,39,43,52 Moreover, despite numerous recent reports in which successful endoluminal stenting for complex posterior circulation aneurysms has been documented,1,9,12,26,28,32,35,43,44,52,59 long-term follow-up data are not yet available and limitations of this therapy have not been fully elucidated. As such, current treatment approaches for these lesions often rely on hunterian strategies with or without cerebral revascularization.6,7,13,14,18,23,25,27,37,38 Segmental isolation of the lesion through trapping, either surgically or by endovascular means, is the treatment of choice,21,27,38 because it definitively uncouples the aneurysm from the intracranial circulation, thereby preventing further growth and the risk of future hemorrhage.

The treatment of dissecting VA pseudoaneurysms involves the origin of the PICA with a trapping procedure poses particular challenge, because PICA revascularization is generally required to avoid brainstem and cerebellar ischemia.5,7,17,23,25,27,37,38,40,42 Several different approaches to PICA revascularization have been described in the neurosurgical literature,21,27,28,32,35,43,46,51 the least common of which is PICA reimplantation (Table 1).21,25,27,38 In this paper, we report on the case of a pediatric patient with a ruptured traumatic VA pseudoaneurysm from which the PICA emerged. The patient was treated with surgical trapping of the aneurysm and direct reimplantation of the PICA origin distal to the diseased vessel segment, resulting in an excellent clinical and angiographically documented outcome. Based on a review of the literature, we are aware of only two other previously documented cases of direct PICA reimplantation in the treatment of complex VA aneurysms:17,38 only one of which involved a VA–PICA anastomosis17 and neither of which occurred in a child. In addition to reporting this extremely rare case and discussing features of commonly used PICA revascularization strategies, we briefly review the existing literature in which PICA reimplantation has been performed as an adjunctive measure in the treatment of complex VA aneurysms.

Case Report

Clinical Presentation. This 13-year-old boy with no significant medical history sustained an accidental gunshot wound through the mouth. He was transported to our insti-
tion where he was initially evaluated by the trauma surgery service and underwent hemodynamic stabilization. Neurological examination revealed the patient to be awake, alert, and with only mild left upper-extremity weakness. No missile exit point was localized. The results of a CT scan of the head were normal; however, imaging of the cervical spine revealed a large bullet fragment lodged in the posterior aspect of the craniovertebral junction and a fracture of the posterior C-1 arch (Fig. 1). The patient was placed in a rigid external orthosis and was then taken to the neuroangiography suite to be evaluated for a VA injury because of the proximity of the bullet fragment to the distal transverse foramen.

Endovascular Procedure. The patient was intubated and general anesthesia was induced. He was prepared and draped in the usual sterile fashion, and his aortic arch was then accessed through the transfemoral route by using a No. 5 French Weinberg catheter and the standard Seldinger technique. Contrast injection of the left VA revealed complete occlusion of this vessel at the level of C-1 (Fig. 2 left). Contrast injection of the right VA, however, demonstrated brisk filling of the BA circulation as well as retrograde flow down the confluence of the sinus, ultimately feeding into a prominent left PICA–AICA complex (Fig. 2 right). There were no noted arterial abnormalities involving the right VA.

Using roadmap guidance, a Renegade microcatheter (Boston Scientific/Target Therapeutics, Inc., Natick, MA) was advanced to the distal left VA immediately proximal to the point of occlusion at the bullet fragment. A total of seven nonretrievable Vortex platinum microcoils (Boston Scientific/Target Therapeutics, Inc.) were then placed in the vessel. After dense coil packing was achieved, the microcatheter was withdrawn. A microcatheter guided by biplane digital subtraction angiography revealed persistent occlusion of the left VA. There was no compromise of flow through the anterior spinal artery or BA.

Postprocedure Course. The patient remained neurologically stable following the coil embolization procedure, and he gradually improved from the bullet injury over the ensuing several days. On Day 5 following his injury, the patient was discharged to a nearby rehabilitation facility to undergo physical and occupational therapy.

On Day 8 following the gunshot wound, the patient developed a significant neurological change with altered mental status and a fixed gaze. He was immediately transferred back to our institution where he complained of a severe headache on admission. Neurological examination revealed the patient to be confused and not able to follow simple commands. A stable, yet persistent, left upper-extremity weakness was demonstrated. A noncontrast CT scan of the head revealed diffuse SAH, intraventricular hemorrhage, and hydrocephalus (Fig. 3 left). The patient was then electively intubated and a right frontal ventriculostomy catheter was inserted. He was taken back to the neuroangiography suite for further evaluation of his intracranial vasculature.

A right VA injection again revealed retrograde flow across the confluence into the distal left VA, with irrigation of a large PICA–AICA complex, the dominant arterial supply to the left side of his cerebellum and brainstem. Just proximal to this major branch, however, there was recanalization of the distal stump and a pseudoaneurysm (Fig. 3 center and right). After considering multiple options, both surgical and endovascular, it was decided to explore the

| TABLE 1 |
| Complex VA aneurysms treated with hunterian strategy and PICA reimplantation* |
| Authors & Year | Hunterian Strategy | Method of PICA Revascularization |
| Durward, 1995 | trapping | direct VA–PICA anastomosis |
| Hamada et al., 1996 | proximal ligation | VA–PICA anastomosis, STA IPG |
| Lee & Sekhar, 1996 | trapping | direct VA–AICA anastomosis |
| Hamada et al., 2002 | trapping | VA–PICA anastomosis, STA IPG |

* IPG = interposition graft; STA = superficial temporal artery.
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area surgically with the goal of placing a clip proximal to the PICA, thereby trapping the aneurysm and preserving blood flow to the left cerebellum and brainstem.

Operation. The patient was placed in the lateral park-bench position and a lateral suboccipital craniotomy was performed. A standard amount of cerebellar retraction was required to expose the pertinent neurovascular anatomy. The ventricular system was decompressed prior to craniotomy by using external drainage. On exposure of the diseased VA segment, it became clear that the PICA origin was actually at the dural insertion of the VA. Following the VA distally approximately 1.5 cm, the aneurysm rupture site was clearly identified. A brainstem perforating branch was seen to emerge from the VA just distal to the aneurysm segment.

Several revascularization options were considered and, because of the vascular anatomy, we decided to reimplant the PICA origin just distal to the aneurysm segment. To this end, burst suppression was initiated in the patient with thiopental and a permanent clip was placed just distal to the aneurysm segment. Appropriately sized temporary clips were applied across the brainstem perforating vessel and distal VA. At its first site of adherence to the brainstem from perforating vessels, a temporary clip was applied to the PICA. The vessel was then severed from the

Fig. 2. Angiograms. Injection of the left VA (left) revealing occlusion at the level of C-1. Injection of the right VA (right) demonstrating brisk filling of the BA as well as retrograde flow down the confluence into a large PICA–AICA complex.

Fig. 3. Noncontrast CT scan of the head (left) revealing diffuse SAH, intraventricular hemorrhage, and hydrocephalus. Anteroposterior (center) and lateral (right) views of the posterior circulation after injection of the right VA demonstrating recanalization of the distal left VA stump and a new pseudoaneurysm.
trapped VA segment and reanastomosed to the VA distal to the aneurysm segment by using 10-0 monofilament nylon suture in an end-to-side fashion. An illustration of the microvascular procedure is depicted in Fig. 4.

After removal of the temporary clips, a moderate amount of perianastomotic hemorrhage was encountered, which was alleviated with gentle tamponade. Brisk pulsatile perfusion of the PICA–AICA complex was visualized all the way to the midline. After achieving hemostasis, the field was irrigated with papaverine, and the wound was closed in the usual fashion.

Postoperative Course. The patient awoke from surgery moving all extremities and following simple commands. A postoperative angiogram revealed clip occlusion of the distal left VA and a functional anastomosis with excellent perfusion (Fig. 5). The patient’s level of mentation gradually improved over the next several days, and he was extubated on postoperative Day 5. On postoperative Day 8, the patient worked closely with staff from physical and occupational therapy. Transient swallowing difficulty, which occurred early in his postoperative course, was absent at the time of discharge 13 days after surgery. He was seen in clinic 1 year later and was found to be neurologically intact. Repeated angiography at that time revealed no significant change from the previous postoperative study.

Discussion

Because of the protective osteofibrous confines in which it resides injury to the VA following penetrating craniocervical trauma is rare.1,2,19,20,28,36,50,56,58 When injury to this vessel occurs, however, structural vascular sequelae can include, but are not limited to, thrombosis, laceration, transection, and traumatic pseudoaneurysm formation.1,2,19,50 False aneurysms, which can arise in an acute or delayed fashion,2,19 occasionally follow a benign course with gradual reconstitution of the normal arterial lumen over time.2 Alternatively, SAH may occur, an event which results in a markedly more ominous natural history.27,33,34,41,45,48,63 In fact, in their account of 42 patients with ruptured pseudoaneurysms of the intracranial vertebrobasilar artery, Mizutani and colleagues46 found a 60% incidence of rebleeding, 57% of which took place during the first 24 hours after the initial hemorrhage. Moreover, each new hemorrhagic episode was associated with a 46% risk of mortality. Because of the grave natural history in the absence of treatment, prompt and definitive intervention is imperative once aneurysm rupture has occurred.

With the advent and subsequent refinements in endovascular techniques, several strategies are currently available for the management of dissecting VA aneurysms. Conceptually, therapeutic modalities are dichotomously categorized as being reconstructive, involving VA preservation, or occlusive, and predicated on VA sacrifice.12,22,23–25,27,38,39,55 Aneurysms that are clearly proximal to the lesion with respect to the origin of the PICA—occlusional therapy. Transient swallowing difficulty, which flow is transmitted into the PICA. For lesions distal to the PICA origin, retrograde perfusion across the cho- roidal point, reimplantation of the PICA in the distal VA supplies the PICA territory.5–7,17,23–25,27,37,38,40,42,46 One approach involves using the OA as a donor pedicle and per- vascularize the PICA territory.5–7,17,23–25,27,37,38,40,46 One approach involves using the OA as a donor pedicle and per- vascularize the PICA territory.5–7,17,23–25,27,37,38,40,46 One approach involves using the OA as a donor pedicle and per- vascularize the PICA territory.5–7,17,23–25,27,37,38,40,46 One approach involves using the OA as a donor pedicle and per- vascularize the PICA territory.5–7,17,23–25,27,37,38,40,46 One approach involves using the OA as a donor pedicle and per- vascularize the PICA territory.5–7,17,23–25,27,37,38,40,46 One approach involves using the OA as a donor pedicle and per- vascularize the PICA territory.5–7,17,23–25,27,37,38,40,46 One approach involves using the OA as a donor pedicle and per-
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or distal to the PICA can usually be trapped without the need for revascularization, provided enough flow is carried through collateral pathways to sustain the basilar circulation. In theory, the optimal method of assessing the presence of sufficient collateral flow is through a formal trial balloon occlusion test including hypotensive challenge. However, in practice, the duplicity of the VAs often permits straightforward angiographic determination. The location of the trapped VA segment with respect to the PICA origin clearly dictates the direction in which flow is transmitted into the PICA. For lesions distal to the PICA origin, orthograde input from the ipsilateral VA supplies the PICA. By contrast, for aneurysms proximal to the PICA origin, retrograde perfusion across the confluence from the contralateral VA subserves the PICA. When the PICA’s origin is from the aneurysm itself—the most complex scenario—trapping should also be performed; however, because of the numerous perforating vessels that arise from the PICA proximal to the choroidal point, definitive revascularization is often required to prevent brainstem and cerebellar ischemia. In some circumstances, patients can tolerate trapping of these aneurysms without PICA revascularization. Certain factors may suggest that proximal PICA sacrifice will be tolerated; however, no steadfast criteria, angiographic or otherwise, are known to exist. Trial balloon occlusion of the PICA origin can be performed to assess such tolerance, although this test is not without limitations. In patients who do not clearly pass trial PICA occlusion or in whom balloon test occlusion is not performed, it has been our preference to err on the side of safety and to perform revascularization.

Several different approaches have been described to revascularize the PICA territory. One approach involves using the OA as a donor pedicle and performing an end-to-side anastomosis with the PICA at an acceptable recipient site. This procedure, although commonly performed, can often be difficult because of early branching of the OA and its rapid loss of caliber in view of the length needed for this deep anastomosis. Moreover, the OA is embedded below a deep fascial layer and is intimately invested within neighboring soft-tissue structures, a feature which necessitates meticulous and time-consuming pre–bypass dissection. Another frequently described approach to PICA revascularization is an anastomosis with its contralateral PICA counterpart. This technique is anatomically appealing as the two vessels come to the midline together in the cisterna magna; however, construction of a durable anastomosis with adequate flow capability requires a healthy contralateral PICA, a feature not always present.

In 1995, Durward was the first to describe PICA reimplantation for the treatment of a ruptured dissecting VA pseudoaneurysm from which the PICA took origin. After trapping the lesion, he performed a direct end-to-side anastomosis between the PICA and VA, proximal to the site of dissection. The following year, Hamada and colleagues reported a case in which they transposed the PICA from a thrombosed giant VA aneurysm to a site on the VA proximal to the lesion. In performing the anastomosis, unlike Durward, these authors used a superficial temporal artery interposition graft. These authors have subsequently expanded this report to include other cases in which the same approach was used. Lee and Sekhar reimplanted the PICA into the ipsilateral AICA in an end-to-side fashion in the treatment of an unreconstructable fusiform aneurysm of the nondominant VA; they achieved an excellent overall outcome. Interestingly, PICA reimplantation in the treatment of a complex VA aneurysm, with or without the use of an interposition construct, has never before been described in a pediatric patient.

Several options for PICA revascularization were considered in our patient. A side-to-side anastomosis between the two PICAs was not selected because the contralateral PICA was, in fact, an AICA–PICA complex, and the PICA branches at the caudal loop were trivial in size. An OA–
PIA bypass was also suboptimal because a preoperative external carotid artery injection revealed a poor caliber donor vessel with early branching. A side-to-side anastomosis of the ipsilateral PICA to the VA at its superior loop was also strongly considered; however, the vessel stretch required was not acceptable. With all other cerebral revascularization options exhausted, reimplantation of the PICA distal to the aneurysm segment was performed with excellent technical, angiographically documented, and clinical success.

Despite the favorable outcome in our patient, one must wonder whether an angiogram, if obtained prior to his initial discharge, would have demonstrated the pseudoaneurysm. Such a finding would have clearly facilitated the lesion being treated prior to SAH, thereby averting the significant potential risks of a hemorrhagic event. At the time the injured left VA was therapeutically coil occluded, a microcatheter run demonstrated excellent coil packing of the lesion and absolutely no antegrade or retrograde flow into the injured vessel segment. We had no reason to suspect that the endoluminal construct would break down and a pseudoaneurysm would arise. That being said, selective post-treatment angiography may have been warranted to ensure a stable result in this case because of the tenuous state of a traumatically injured intracranial vessel, the known limitation of proximal occlusion, and the dangerous natural history of these lesions once aneurysm rupture has occurred.

Conclusions

Dissecting intracranial VA pseudoaneurysms involving the PICA origin are uniformly challenging lesions with a potentially dismal natural history once rupture has occurred. Trapping of the aneurysm segment often requires PICA revascularization to avoid brainstem and cerebellar ischemia. Of the many methods of PICA revascularization, reimplantation into the VA is a safe and effective strategy and may be successfully performed in the pediatric population.

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