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Case Report

# Ruptured proximal pontine artery aneurysm and association with cerebellopontine angle cistern arterial venous malformation fed by the same artery: A surgical challenge

Maurizio Gladi<sup>1</sup><sup>(b)</sup>, Alessandro Di Rienzo<sup>1</sup>, Maria Rossella Fasinella<sup>1</sup>, Denis Aiudi<sup>1</sup>, Paolo Balercia<sup>2</sup>, Mauro Dobran<sup>1</sup>, Maurizio Iacoangeli<sup>3</sup><sup>(b)</sup>

<sup>1</sup>Department of Neurosurgery, Università Politecnica Delle Marche Facoltà di Medicina e Chirurgia, <sup>2</sup>Department of Maxillo-Facial Surgery, Ospedali Riuniti di Ancona, <sup>3</sup>Department of Neurosurgery, Università Politecnica delle Marche, Ancona, Italy.

E-mail: Maurizio Gladi - mauriziogladi@gmail.com; Alessandro Di Rienzo - alessandrodirienzo1@gmail.com; \*Maria Rossella Fasinella - rossella.fasi@gmail.com; Denis Aiudi - denis.aiudi@gmail.com; Paolo Balercia - paolo.balercia@ospedaliriuniti.marche.it; Mauro Dobran - dobran@libero.it; Maurizio Iacoangeli - neurotra@gmail.com

#### \*Corresponding author:

Maria Rossella Fasinella, Department of Neurosurgery, Università Politecnica Delle Marche Facoltà di Medicina e Chirurgia, Ancona, Italy.

#### rossella.fasi@gmail.com

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# ABSTRACT

**Background:** The coexistence of hyper-inflow aneurysms and cerebellopontine angle cistern (CPAc) arterial venous malformations (AVMs) have been rarely reported and most commonly associated with high risk of bleeding.

**Case Descriptions:** We present two cases of CPAc AVMs admitted for acute subarachnoid hemorrhage from rupture of a parent right pontine artery aneurysm. Admission history, neurology at presentation, pre/post-operative imaging, approach selection, and results are thoroughly reviewed and presented. The acute origin angle of the vessel from the basilar artery made both malformations unsuitable for endovascular treatment. The surgical strategy was differently tailored in the two patients, respectively, using a Le Fort I/transclival and a Kawase approach. The aneurysm was clipped in the first case, and the AVM was excised in the second one, as required by the anatomical context. Aneurysm exclusion and AVM size reduction were obtained in the first case, while complete AVM removal and later aneurysm disappearance were obtained in the second one. A high-flow cerebrospinal fluid leak in the first case was successfully treated by an endoscopic approach. Both patients experienced a satisfactory neurological outcome in the follow-up.

**Conclusion:** Pontine artery aneurysms, especially when associated with CPAc AVMs, represent a surgical challenge, due to their rarity and anatomical peculiarity, which typically requires complex operative approaches. Multimodal preoperative imaging, appropriate timing, and accurate target selection, together with versatile strategies, are the keys to a successful treatment.

Keywords: Aneurysm, Arteriovenous malformation, Cerebellopontine angle, Clipping, Pontine artery

# INTRODUCTION

Only 8–12% of intracranial aneurysms and 5–15% of arterial venous malformations (AVMs) occur in the posterior circulation.<sup>[27,29]</sup> Cerebellopontine angle cistern (CPAc) AVMs are even rarer,<sup>[32]</sup> especially when associated with aneurysms originating from the same parent artery

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(2.8–9.3% of all AVMs).<sup>[25]</sup> Posterior fossa AVMs also have a documented tendency to develop aneurysms compared to their anterior circulation counterparts.<sup>[29]</sup> The theoretical basis for this event stays in the increased blood flow coming through high-pressure vessels, a major factor in aneurysmal genesis.<sup>[2,6]</sup> Other suggested hypotheses may implicate either the decreased resistance inside AVM nidus<sup>[23]</sup> or the increased hemodynamic turbulence in feeding arteries,<sup>[36]</sup> though the contribution of both factors remains unclear.

The categorization of these aneurysms is based on their relationship with the nidus, extranidal ones more frequently occur on the pedicle of the feeding artery and are considered the leading cause of bleeding in about 37% of cases.<sup>[29,37]</sup> Either microsurgical or endovascular procedures, frequently combined, are suitable for such an aim.<sup>[3,14,22,26,30]</sup> However, the major challenge in management remains the proximity of these complex malformations to vital brainstem structures.<sup>[20,24,33]</sup> In this context, we present the operative management of two CPAc AVMs – pontine artery aneurysms, both supplied by the same feeder artery, characterized by a small AVM nidus and a proximal ruptured aneurysm.

# CASE DESCRIPTIONS

# Illustrative case 1

A 63 year old patient was admitted for acute onset of nuchal headache, vomiting, and dysarthria. The emergency head computerized tomography (CT) scan showed the presence of subarachnoid and intraventricular hemorrhage, predominantly located inside the right CPA and prepontine cisterns. The following CT-angiography (CTA), completed with magnetic resonance angiography (MRA) and digital subtraction angiography (DSA), disclosed the presence of a small right-sided CPAc AVM, with the ipsilateral dilation of a parent pontine artery and a hyper-inflow aneurysm (maximum diameter 4.5 mm), deep-seated in the brainstem, at its takeoff from the basilar artery (BA). A second aneurysm was also appreciated distally to the nidus. AVM's venous drainage was supplied from a right anterolateral pericerebellar vein. The proximal aneurysm was recognized as the leading source of bleeding, and during the following 3 weeks, its size progressively increased to reach a diameter of 6.5 mm at followup DSAs [Figure 1]. Due to the sharp angle origin of the parent artery from the basilar trunk and a documented vasospasm, two attempts at endovascular treatments were unsuccessful. To address the source of bleeding and considering the small size of the AVM, we decided to clip the aneurysm and refer the residual AVM to Gamma Knife Surgery (GKS).

# Surgical technique

Due to the aneurysm location, a straightforward approach through a median anatomical trajectory was selected. In

this context, the transmaxillary Le Fort type I approach was performed, in cooperation with the head and neck surgeons. The oral mucosa and muscles were elevated from the maxilla superiorly, up to the infraorbital nerve. After exposing the piriform opening, the nasal mucosa was stripped, reaching the level of the inferior turbinates. A horizontal osteotomy was made from the piriform opening through the maxillary alveolus using a combination of oscillating saw and piezoelectric osteotome. The maxilla was disarticulated and down-fractured by inserting a chisel and applying downward pressure. The inferior turbinates were fractured and removed with rongeurs. With a tongue retractor holding in place the transected maxilla, the transoral retractor was placed against the palatal plate. The nasal mucosa was reflected off the septum in order to expose the sphenoid base. The clival/paraclival region was finally exposed through a posterior mid-pharyngeal incision. After this step and under navigation guidance, we performed a targeted opening of the sellar floor and clivus, using a combination of drilling and piezosurgery, exposing both the carotids and the posterior clinoid processes. A targeted dural opening led to the identification of the basilar trunk, the parent artery, and the aneurysm. Combining microsurgical and endoscopic techniques, a slightly curved clip was applied, reaching full aneurysm exclusion. The vessel's patency was verified with a Doppler probe and Indocyanine Green. Adipose tissue was harvested from the abdominal region to close the dural defect and fill the entire sphenoidal sinus. Part of the septum cartilage was used to strengthen the closure, finally reinforced with fibrin glue. The split bone was fixed and reapproximated with titanium mini-plates.

Postoperative DSA confirmed aneurysm exclusion and the parental artery was not appreciable, supposedly due to a stenosis of the vessel itself. Reduced inflow to the distal AVM nidus was still visible, due to retrograde supply from an ipsilateral superior cerebellar artery (SCA) and anterior inferior cerebellar artery (AICA) [Figure 2]. One week later, the patient presented a high-flow cerebrospinal fluid (CSF) leak and underwent an endoscopic revision. The patient was discharged home with no neurological disturbances and underwent GKS 2 months later. He is actually awaiting the follow-up DSA.

# Illustrative case 2

A 48-year-old man admitted to our emergency room with severe nuchal headache and no neurological focal deficits underwent a CT scan showing a right cerebellopontine and prepontine cisterns subarachnoid hemorrhage (SAH). The following CTA and MRA were suggestive of a CPAc AVM. A DSA documented a small-sized right CPAc AVM originating from a fusiform dilation at the origin of the pontine artery, the presumed source of the bleeding



**Figure 1:** (a) Admission computerized tomography (CT) scan showing subarachnoid hemorrhage involving the preportine and cerebellopontine cisterns; (b) From the left: CT-angiography coronal view showing a right pontine artery aneurysm (red arrow); magnetic resonance angiography confirming the known aneurysm (red arrow) and a cerebellopontine angle cistern arterial venous malformation (white arrow) (c) admission (left) and 3 weeks (right) digital subtraction angiography demonstrating aneurysm growth from 4.5 mm to 6.5 mm.

[Figure 3]. Endovascular treatment of both lesions failed due to proximal stenosis of the parent artery, making the vessel unsuitable for catheter navigation. Due to the fusiform shape of the aneurysm, we decided to target first the AVM through a Kawase approach. The goal of the surgical procedure was to reduce the flow either in the parent artery or in the aneurysm. Preoperatively, an external lumbar drain was positioned.

#### Surgical technique

The patient was placed supine with the head turned left. A right subtemporal approach was performed in the standard fashion. The zygomatic arch was exposed, preplated, and removed to pull down the temporalis muscle and gain straight access to the middle cranial fossa. After extradural dissection and CSF subtraction, the bony markers of Kawase's

quadrangle were identified and the petrous apex was drilled. After dural incision and tentorial exposure, the IV<sup>th</sup> cranial nerve (CN) was identified and the tentorium cut behind it to visualize the posterior cerebral artery, III<sup>rd</sup> CN, and SCA, up to the VII<sup>th</sup>–VIII<sup>th</sup> CNs. The lateral surface of the brainstem and the AVM was seen. The nidus was isolated and removed after an endoscopic exploration. Finally, a layer-by-layer reconstruction of the middle fossa floor was performed to prevent CSF leakage. The zygomatic arch was finally reapposed.

In the immediate postoperative period, the patient experienced transient diplopia due to a stupor of the VI<sup>th</sup> right CN, fully resolved 4 weeks later. One week postoperative, DSA confirmed AVM exclusion and downsizing of the aneurysm. Six months later, the aneurysm itself was no longer visible [Figure 4].



**Figure 2:** (a) To the left early mid-facial degloving with piezosurgery, in the middle drilling of the clivus (c), and to the right position check under neuronavigation; (b) aneurysm (A) exposure. The brainstem (B) is visualized beyond the aneurysm dome, final clip position, and (right) closure; (c) computerized tomography bone window demonstrates a tailored bone removal and position of the clip (white arrow). Reduced inflow to the distal AVM nidus is still visible (red arrow). Anterior-posterior angiographic view shows clipping of the aneurysm, which appears completely excluded, as the course of the pontine artery itself due to vessel stenosis. To the right final cosmetic result.

#### DISCUSSION

Posterior fossa AVMs associated with flow-related aneurysms carry a significantly higher rate of rupture and re-bleeding if compared to the isolated aneurysm counterparts.<sup>[28]</sup> When the blood flow increases in the AVM feeding vessel, it predisposes to aneurysm development. This seems to be supported by reports of spontaneous aneurysm disappearance after AVM removal.<sup>[14,35,36]</sup> If the treatment choice consists of targeting the aneurysm or AVM alone, the risk of the residual lesion to bleed remains, due to regional hemodynamic changes.<sup>[31,39]</sup> Under these circumstances, Cunha e Sa *et al.* recommended that the symptomatic lesion, if determined, should be treated first.<sup>[7]</sup>

Therapeutic intervention for AVMs with associated prenidal aneurysms are complex and multifaceted. As endovascular treatment options have evolved to reduce surgical-related morbidity, the management of posterior circulation aneurysms has significantly changed.<sup>[25]</sup>

In our first described case, the endovascular option of a glue embolization was too dangerous due to the absence of a reflux margin, resulting in the risk of distal glue embolization in basilar territory; in the second one, coiling also turned out to be unfeasible in view of the tortuosity of the origin angle of the artery from the basilar trunk, which made the maneuvering space extremely narrow to navigate the pseudoaneurysm and to catheterize the vessel distally to the aneurysm.

GKS is a treatment option for AVMs, but its effect is not immediate and the risk of re-bleeding may persist for up to 2 years.<sup>[20,22]</sup>

To the best of our knowledge, this paper is the first report of two surgically treated patients with proximal pontine artery aneurysms and association with CPAc AVM fed by the same artery. Three more cases were reported in the literature and the onset sign was always SAH. One patient was addressed to



**Figure 3:** (a) Computerized tomography (CT) and axial magnetic resonance imaging showing basal cisterns subarachnoid hemorrhage (white arrow); (b) from the left: CT-angiography and magnetic resonance angiography (axial view). Aneurysm (black arrow) and arterial venous malformation (AVM) (white arrow); (c) Digital subtraction angiography (anterior-posterior view) outlines the specific characteristics of the vascular malformation; small right cerebellopontine angle cistern AVM (red arrow) with proximal aneurysm (white arrow) whose origin was from a dilated pontine artery. Other minor feeders to the AVM seem to arise from the ipsilateral superior cerebellar artery and anterior inferior cerebellar artery.

GKS, with a residual nidus reduced in size after 16 months. In the remaining two, a combination of aneurysm embolization and AVM radiosurgery was used.<sup>[28,30,32]</sup>

In our cases, the aneurysms were both placed at the junction between the BA and a dilated pontine artery. This, further, supports the concept that similar lesions might be flowrelated rather than linked to an intrinsic vessel wall alteration. A key concept to consider is that both lesions, AVM and aneurysm, are equally exposed to the same high-pressure arterial flow simultaneously since they recognize the same arterial origin.<sup>[21]</sup>

The treatment of posterior circulation aneurysms includes limited surgical options, due to the narrow angles of approach and the overall difficulty in ensuring adequate proximal control. Microsurgical clipping of pontine artery aneurysms is especially challenging due to their location within the prepontine cistern, requiring penetration along a deep and narrow surgical corridor,<sup>[36]</sup> in contiguity to critical neurovascular structures.

Several skull base approaches, including the far-lateral, retrosigmoid, presigmoid (with different degrees of petrosectomy), subtemporal, anterior transpetrosal, orbitozygomatic, and trans-oral/facial/clival,<sup>[12,13,27]</sup> have been proposed to address these lesions. Unfortunately, they are all affected by insufficient lightning, limited exposure of perforating arteries, difficult proximal control, and dissection of the aneurysm, with consistently raised operative risks. Lawton *et al.*<sup>[24,36]</sup> reported up to a 60% complication rate.

Since the choice of the approach is of paramount importance, two factors seem to be strategical to this aim: (1) the craniocaudal location of the aneurysm in relation to the clivus and (2) the mediolateral extension in relation to the course of the artery. The relative position of the VI<sup>th</sup> CN with respect to the aneurysm may play a role itself in this choice.<sup>[1]</sup>

First reported by Sano in 1979, the transclival route to the BA for clipping of cerebral aneurysms was subsequently described by other groups.<sup>[36]</sup> Early authors used either the transoral transpalatal<sup>[8]</sup> or the transcervical transclival approaches,<sup>[36]</sup> both associated with major morbidities. Crockard et al. were the first to describe the successful transoral clipping of a basilar trunk aneurysm.<sup>[5]</sup> This report underlined how palatal splitting could provide adequate exposure, even with the help of a new long-handled applier for the clip. The transoral transpalatal approach may also furnish a similar surgical exposure, but palatal dysfunction may follow, potentially resulting in voice change, dysphagia, nasal regurgitation, and oronasal fistulas.<sup>[5,38]</sup> Through the transfacial transclival route, dural exposure from the sellar floor to the anterior foramen magnum can be achieved, while the distance from the midline remains the main anatomic limitation of such an approach due to the lateral presence of the internal carotid arteries.[11,17] Ogilvy successfully clipped five aneurysms through the transfacial transclival route,<sup>[34]</sup> despite high rates of CSF leak and meningitis secondary to difficulties in the reconstructive phase. It was unclear if clip presence within the clival defect might affect dural repair efficacy.<sup>[16]</sup> On the other hand, the technique provided a satisfactory anterior view of the prepontine cistern and early exposure of the BA, neck of the aneurysm, and parent artery, ensuring safe aneurysm dissection, good proximal control, and minor risk of the lower CNs dysfunction. To confirm this observation in the first of



**Figure 4:** (a) Exposure of the Kawase quadrangle and anterior petrosectomy performed by the use of the piezoelectric osteotome and drilling. On the right, endoscopic close-up view of the arterial venous malformation (AVM) (white arrow); (b) AVM coagulation under microscope and, on the right, final endoscopic assessment of complete AVM exclusion (white arrow); (c) postoperative studies. A baseline computerized tomography scan shows the right anterior petrosectomy. Digital subtraction angiography (anterior-posterior view) performed after 7 days showed complete AVM removal and dimensional reduction of the proximal pontine artery aneurysm. To the right 6-months, magnetic resonance angiography detected no residual vascular anomalies.

our cases, the paramedian position of the aneurysm, deeplocated within the brainstem, perfectly fits the anterior access strategy. With the above-listed limitations, some difficulties were still found in identifying the lesion, dissecting it, and positioning the clip without pushing against the brainstem. The approach allowed a straight, nonobstructed route to the aneurysm, and without exposure and manipulation of other critical neurovascular structures.

Laterally directed approaches were first popularized by Drake,<sup>[8]</sup> for the exposure of 32 AICA aneurysms, but the visualization of the midline was limited. In addition, the drilling of the petrous apex in the subtemporal transtentorial corridor may jeopardize the IV<sup>th</sup> CN during the tentorial edge opening. In the second reported case, we chose this

access, which provided better control of the AVM and both the afferent and efferent vessels, while preserving hearing.

In both patients, the use of endoscopic assistance was crucial to better define locoregional anatomy, highlighting the relationship between the lesion and the vital surrounding structures and to assess the optimal clip placement. Its simultaneous use of microscopic vision was essential to avoid damages related to retraction and unnecessary dissection.<sup>[4,9,10,15,18,19]</sup>

# CONCLUSION

Proximal pontine artery aneurysms and association with CPAc AVMs fed by the same artery are rare and life-

threatening lesions. Choosing the type and timing of treatment is strategic, with surgical options being limited due to the complexity of adjacent neurovascular anatomy. Tailoring treatment is necessary and requires a multidisciplinary team, including neuro, head and neck, and ENT surgeons, interventional neuroradiologists, anesthetists, physiotherapists, and radiosurgeons, to optimize the final outcome.

#### Authors contributions

All authors contributed to the study's conception and design.

#### **Ethical approval**

All procedures performed in studies involving human participants were in accordance with the ethical standards of the Institutional Committee and with the latest amendment of the Helsinki Declaration.

#### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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# **Conflicts of interest**

There are no conflicts of interest.

# Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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