

# ***Persistence of Primitive Lateral Basilovertеbral Anastomosis with a Ruptured Posterior Inferior Cerebellar Artery Aneurysm: A Case Report***

Takahiro OTA,<sup>1</sup> Shogo DOFUKU,<sup>1</sup> and Masayuki SATO<sup>1</sup>

<sup>1</sup>*Department of Neurosurgery, Tokyo Metropolitan Tama Medical Center, Fuchu, Tokyo, Japan*

## **Abstract**

**Posterior inferior cerebellar artery (PICA) variations are well recognized; however, their mechanisms have not been well understood. Primitive lateral basilovertеbral anastomosis (PLBA) was described in 1948 by Padget and is one of the embryological transient longitudinal channels in the hindbrain. This study reports a PICA aneurysm associated with PLBA. A 48-year-old man presented with subarachnoid hemorrhage. Cerebral angiography showed a 3.6-mm fusiform aneurysm with a bleb of the left PICA just at the origin of the PICA from the vertebral artery. Furthermore, a direct anterior inferior cerebellar artery (AICA)-PICA anastomosis parallel to the basilar artery was revealed. This direct anastomosis between the AICA and PICA is explained by the partial persistence of PLBA. Variations in the three cerebellar arteries and vertebrobasilar junction can be caused by persistence of PLBA.**

Keywords: primitive lateral basilovertеbral anastomosis, persistence, posterior inferior cerebellar artery, aneurysm, embryology

## **Introduction**

The posterior inferior cerebellar artery (PICA) is one of three long cerebellar arteries that supply the cerebellum. Embryologically, these three transverse arteries from the longitudinal channel enlarge to meet the arterial supply demands of the cerebellum.<sup>1)</sup> The PICA and anterior inferior cerebellar artery (AICA) are in balance with each other due to the need of blood supply by the cerebellum. Usually, in patients without a PICA, the ipsilateral AICA supplies blood to the posteroinferior part of the cerebellum.

Primitive lateral basilovertеbral anastomosis (PLBA) in humans was described in 1948 by Padget as an embryological longitudinal channel that links the developing lateral branches of the vertebrobasilar system.<sup>2)</sup> Gregg et al. explained the common trunks of origin and aberrant origins of the three cerebellar arteries by partial persistence of this anastomosis.<sup>3)</sup>

Herein, we report a rare anastomosis between the AICA and PICA with ruptured PICA aneurysm, which was explained by the partial persistence of PLBA.

## **Case Report**

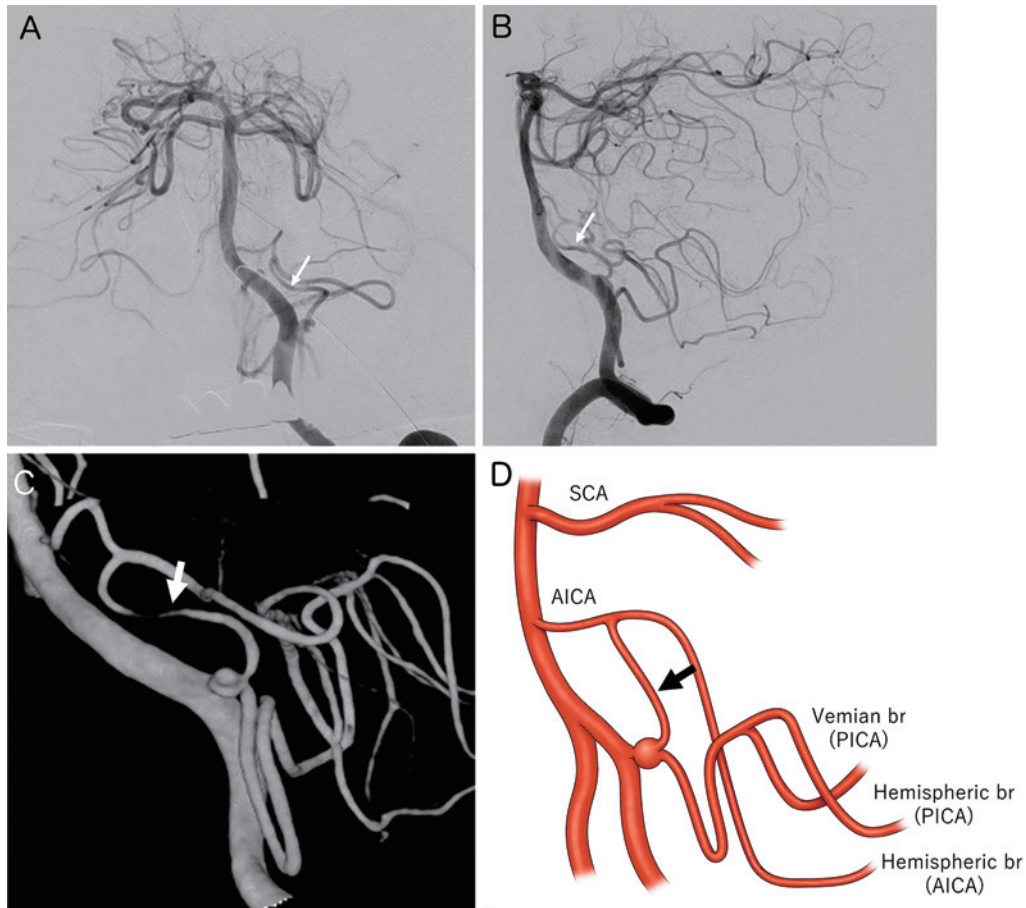
A 48-year-old man presented with a sudden conscious disturbance. His Glasgow Coma Scale score was E1V1M1, blood pressure was 118/77 mmHg, and pupil size was 4 mm on the right side and 3 mm on the left side. Head computed tomography revealed thick subarachnoid hemorrhage, particularly in the left cerebellomedullary cistern. Cerebral angiography showed a 3.6-mm fusiform aneurysm with a bleb of the left PICA right at the origin of the PICA from the vertebral artery (VA). Furthermore, an AICA-PICA anastomosis parallel to the basilar artery (BA) was revealed. Three-dimensional (3D) rotational angiography showed that the AICA has two branches: the AICA-PICA anastomosis and the hemispheric branch, with the PICA running along the lateral side of the medulla and bifurcating into the hemispheric and vermian branches (Fig. 1).

Emergency craniotomy and clipping of the aneurysm were performed on day 0. The aneurysm was exposed via left suboccipital craniotomy after harvesting the left occipital artery in preparation for the occipital artery-PICA bypass. The rupture point was at the bleb of the aneurysm, and no perforators from the proximal segment of the PICA, the dome and neck of the aneurysm, and the AICA-

Received December 1, 2021; Accepted February 10, 2022

Copyright © 2022 The Japan Neurosurgical Society

This work is licensed under a Creative Commons Attribution-NonCommercial-NoDerivatives International License.



**Fig. 1** A, B: Cerebral angiography showing a 3.6-mm fusiform aneurysm with a bleb of the left PICA at the origin of the PICA from the vertebral artery (VA). Additionally, an AICA-PICA anastomosis parallel to the basilar artery (BA) is revealed.

**C:** 3D rotational angiography showing AICA-PICA anastomosis (white arrow).

**D:** Illustration of Fig. 1C. The AICA has two branches: the AICA-PICA anastomosis and the hemispheric branch and the PICA running along the lateral side of the medulla and bifurcating into the hemispheric and vermian branches. (Illustrated by (C) SAIKOU, INC.)

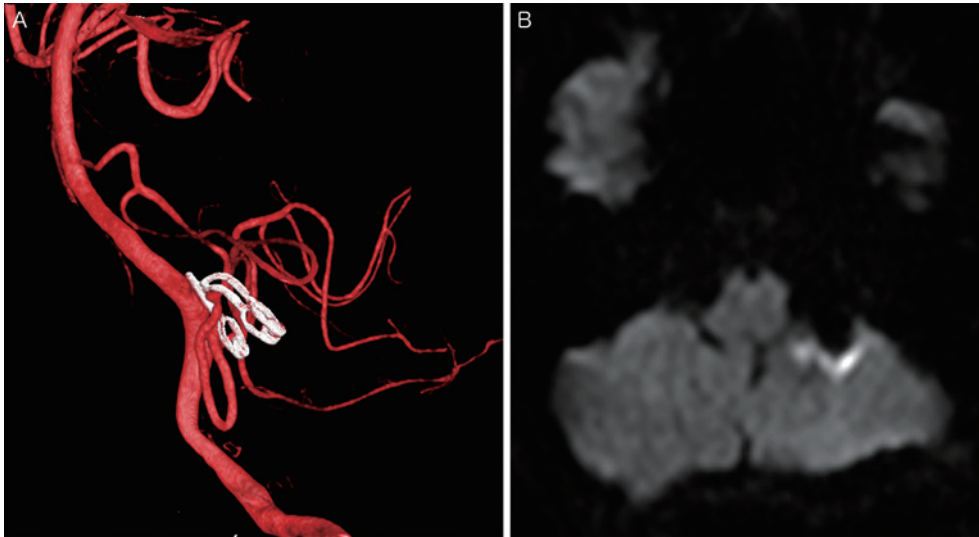
PICA segment were observed in the operative field. Aneurysm trapping was performed using titanium clips, and the vascular supply to the PICA branches was preserved via AICA-PICA anastomosis (Fig. 2A). Eventually, the left AICA supplied the entire inferior cerebellum perfused by both the AICA and PICA. Postoperative magnetic resonance imaging showed no infarction in the brainstem or cerebellum (Fig. 2B), and the patient's neurological condition improved greatly, except for bilateral abducens nerve palsy. He was discharged on day 24 with a modified Rankin scale score of 1. There was no recurrence of the aneurysm or worsening of neurological symptoms after 2 years of postoperative follow-up.

## Discussion

This report presents the case of a patient with persistent PLBA and ruptured PICA aneurysm that was successfully treated. PLBA is a prominent feature at certain stages

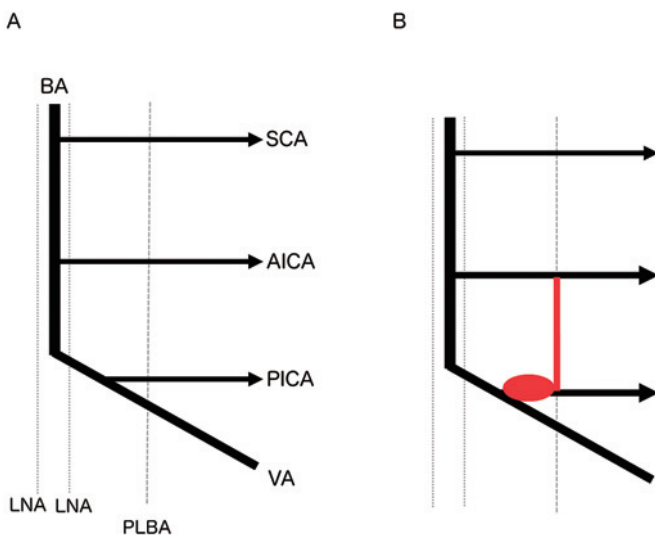
of human hindbrain development. Many studies have reported anatomical variations of the PICA; however, only few case reports on aneurysms related to PLBA,<sup>4,5)</sup> which have discussed PLBA in detail, are available in the literature.<sup>3)</sup> This is likely due to PLBA not being well recognized in the neurosurgical or neurointerventional fields.

Embryologically, the BA is formed from the fusion of longitudinal neural arteries (LNAs). PLBA is a longitudinal anastomosis temporarily present on the hindbrain (including the pons, medulla, and cerebellum) lateral wall. It connects the developing lateral branches of the LNAs during development of blood supply to the hindbrain. PLBA and the LNAs are parallel, and part of PLBA will later form the proximal segments of the three cerebellar arteries.<sup>2)</sup> PLBA is supplied caudally by the posterior branch of the proatlantal artery and cranially by the lateral branches of the LNA (particularly, the primitive trigeminal artery). It exists from an embryo of 5-6 mm ( $28 \pm 1$  days) to 12-14 mm ( $32 \pm 1$  days) of development of the embryonic cerebral arte-



**Fig. 2 A:** Postoperative 3D rotational angiography showing complete obliteration of the aneurysm, with the branches of the AICA and PICA being preserved.

**B:** Postoperative diffusion-weighted image showing no infarction in the medulla or left cerebellum.



**Fig. 3 A:** Normal adult.

**B:** Patient in our study. Partial persistence of PLBA (red line) and aneurysm formation at the origin of the PICA.

LNA: longitudinal neural artery, PLBA: primitive lateral basilo-vertebral anastomosis.

rial system.<sup>2)</sup> In 1957, Moffat noted the lateral longitudinal artery (LLA) in rats, which corresponded to the PLBA observed by Padget in humans.<sup>6)</sup> The LLA in rats is a primary vessel that later acquires anastomoses with the LNAs. The development of PLBA in humans has a close resemblance to that of the LLA in rats, except that in the human embryo, much of the PLBA disappears and some of its dorsal branches keep numerous anastomoses with the PICA.<sup>7)</sup>

Three cerebellar arteries are transverse arteries from the LNAs, and PLBA enlarges to meet the demands of the

cerebellum and brain stem. In the early stage (12-14 mm embryo;  $35 \pm 1$  days), only the superior cerebral artery supplies cerebellar rudiment. The AICA and PICA develop later, and the dominance of either of these arteries is determined by the extent of the cerebellar surface it captures. The posterior inferior portion of the cerebellar hemispheres is vascularized in the late stage; therefore, the late development of the PICA could explain the large variability in its pathway as well as its several deviations.<sup>1)</sup> The double and extradural origins of the PICA are the result of the transfer, regression, and persistence of various arterial segments of the embryonic vertebrobasilar system, including PLBA.<sup>8)</sup> Macchi et al. reported morphogenesis of the PICA using 3D reconstruction<sup>1)</sup> and showed that PLBA is a large channel that constitutes the main vessel on the lateral aspect of the hindbrain. PLBA is an elective candidate for incorporation in the stem of the PICA, with an intriguing chronological correspondence between PICA appearance and PLBA regression at the end of the 8th week.<sup>1)</sup> In our case, the direct connection between the AICA and PICA was located on the lateral wall of the brainstem, so it is reasonable to consider that this segment is derived from the partial persistence of PLBA (Fig. 3).

There are only few reports on aneurysm-related PLBA. Yagi et al. reported a case of subarachnoid hemorrhage in which the BA was supplied primarily by persistent PLBA because of incomplete development of the ipsilateral vertebrobasilar junction.<sup>9)</sup> In their case, the aneurysm was formed at the left VA-AICA contralateral to the BA-PLBA anastomosis joint, which was also attributable to excessive hemodynamic stress via PLBA. Furthermore, Joshi et al. reported a case of BA duplication with an aneurysm, accompanied by persistence of PLBA at the origin of the PICA.<sup>5)</sup>

BA duplication needs to be distinguished from BA fenestration. The role of PLBA in the formation of vertebrobasilar duplications has been previously reported.<sup>3,9)</sup> In vertebrobasilar duplication, the lateral limb is made of a persistent segment of PLBA, whereas the medial limb corresponds to the normal junction.<sup>3)</sup>

This PICA fusiform aneurysm developed at the short PICA between the VA and the site of AICA-PICA anastomosis. The fenestration or duplication is extremely vulnerable to aneurysm formations.<sup>10)</sup> Muscular gaps were documented in the tunica media of the medial walls at the two ends of the fenestration.<sup>11)</sup> The junction of PICA and AICA-PICA anastomosis (PLBA) in our case may have some structural defects. PLBA is a normal variant and has no pathological vulnerability on its own; however, the partial persistence of the PLBA (AICA-PICA anastomosis) complicates the blood flow at the junction to the PICA. We speculate that these muscular gaps and complicated hemodynamic stress due to PLBA acted as factors in the aneurysmal formation in this case.

Most clinicians do not recognize the relationship between various PICA variations and the persistence of PLBA. Therefore, the exact frequency of aneurysm occurrence associated with PLBA is unclear, and it is uncertain whether these aneurysms are easily formed and prone to rupture. As mentioned above, anastomosis and duplication between the AICA and PICA can be explained as partial persistence of PLBA. With the findings of our study, we assume that many more cases associated with PLBA are present than is recognized.

### Acknowledgments

The authors would like to express their special thanks to Dr. Masaki Komiyama, Department of NeuroIntervention, Osaka City General Hospital, for his insightful suggestions and comments.

### Conflicts of Interest Disclosure

The authors report no conflicts of interest (COI) concerning the materials or methods used in this study or the

findings specified in this paper. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

### References

- 1) Macchi V, Porzionato A, Guidolin D, Parenti A, De Caro R: Morphogenesis of the posterior inferior cerebellar artery with three-dimensional reconstruction of the late embryonic vertebrobasilar system. *Surg Radiol Anat* 27: 56-60, 2005
- 2) Padget DH: The development of the cranial arteries in the human embryo. *Contrib Embryol Carnegie Instn* 32: 205-261, 1948
- 3) Gregg L, Gailloud P: The Role of the Primitive Lateral Basilovertbral Anastomosis of Padget in Variations of the Vertebrobasilar Arterial System. *Anat Rec* 300: 2025-2038, 2017
- 4) Yagi K, Satoh K, Satomi J, Nagahiro S: Primitive vertebrobasilar system associated with a ruptured aneurysm. *Am J Neuroradiol* 25: 781-783, 2004
- 5) Joshi KC, Thomas A, Jagannatha AT, Garg D: Stent-Assisted Coiling of Aneurysm in a Persistent Primitive Lateral Vertebrobasilar System. *World Neurosurg* 86: 513.e9-513.e14, 2016
- 6) Moffat D: The development of the hind-brain arteries in the rat. *J Anat* 91: 25-39, 1957
- 7) Moffat D: The embryology of the arteries of the brain. *Ann R Coll Surg Engl* 30: 368-382, 1962
- 8) Lasjaunias P, Vallee B, Person H, Ter Brugge K, Chiu M: The lateral spinal artery of the upper cervical spinal cord. Anatomy, normal variations, and angiographic aspects. *J Neurosurg* 63: 235-241, 1985
- 9) De Caro R, Parenti A, Munari PF: Persistent primitive lateral vertebrobasilar anastomosis. *Acta Neurochir (Wien)* 138: 592-594, 1996
- 10) Sanders W, Sorek P, Mehta B: Fenestration of intracranial arteries with special attention to associated aneurysms and other anomalies. *AJNR Am J Neuroradiol* 14: 675-680, 1993
- 11) De Caro R, Serafini M, Galli S: Anatomy of segmental duplication in the human basilar artery: possible site of aneurysm formation. *Clin Neuropathol* 14: 303-309, 1995

---

Corresponding author: Takahiro Ota, MD, PhD

Department of Neurosurgery, Tokyo Metropolitan Tama Medical Center, 2-8-29 Musashi-dai, Fuchu, Tokyo 183-8524, Japan.

e-mail: takaota@tama-mail.jp