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Case Report

Mechanical Thrombectomy for Basilar Artery Occlusion with a Type 1 Persistent Proatlantal Artery: A Case Report and Literature Review

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Objective: Persistent proatlantal artery (PPA) is a primitive carotid-vertebrobasilar anastomosis (CVA); acute ischemic stroke due to basilar artery (BA) occlusion via a PPA is extremely rare.

Case Presentation: An 84-year-old female developed disturbance of consciousness (Glasgow Coma Scale E2V1M5) and quadriparesis with a National Institutes of Health Stroke Scale score of 35. Head CT revealed early ischemic changes in the right temporal lobe, and a hyperdense vessel sign in the BA. Cerebral angiography showed that the left vertebral artery (VA) did not originate from the left subclavian artery or aortic arch. A left common carotid artery angiogram showed the presence of the left PPA originating from the left external carotid artery. Mechanical thrombectomy (MT) with contact aspiration using a Penumbra 5MAX ACE 60 aspiration catheter was performed, and successful recanalization was achieved after clot retrieval in the first attempt (thrombolysis in cerebral infarction scale 2b). MRI performed the following day, however, revealed a newly developed large hemorrhagic infarction in the pons, with no improvement in her symptoms (modified Rankin Scale score of 5 at 90 days).

Conclusion: Although MT achieved successful recanalization of the BA via the PPA, her clinical symptoms did not improve, probably because of poor collateral circulation or the long length of the occlusion. In patients with acute vertebro-BA occlusion, if the VA does not originate from the subclavian artery or aortic arch, the presence of a primitive CVA should be considered.

Keywords ▶ persistent proatlantal artery, basilar artery occlusion, mechanical thrombectomy, endovascular treatment, acute ischemic stroke

Introduction

Persistent proatlantal artery (PPA), which represents a primitive carotid-vertebrobasilar anastomosis (CVA), is extremely rare, with an incidence of 0.02%.[1)] The PPA typically originates from the common carotid artery (CCA), internal carotid artery (ICA), or external carotid artery (ECA) and connects with the vertebral artery (VA) at the suboccipital space.[2)] Although the PPA is the main blood supply for the vertebrobasilar system in these cases, acute ischemic stroke (AIS) due to basilar artery (BA) occlusion in patients with a PPA is extremely rare, and only three such cases have been reported previously,[3–5)] only one of which was treated by mechanical thrombectomy (MT).[5)] This report presents a case of AIS due to BA occlusion via a PPA who presented with disturbance of consciousness, quadriparesis, and anisocoria, which were treated by contact aspiration thrombectomy.

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Case Presentation

An 84-year-old female who was under medical treatment for acute heart failure at another hospital developed disturbance

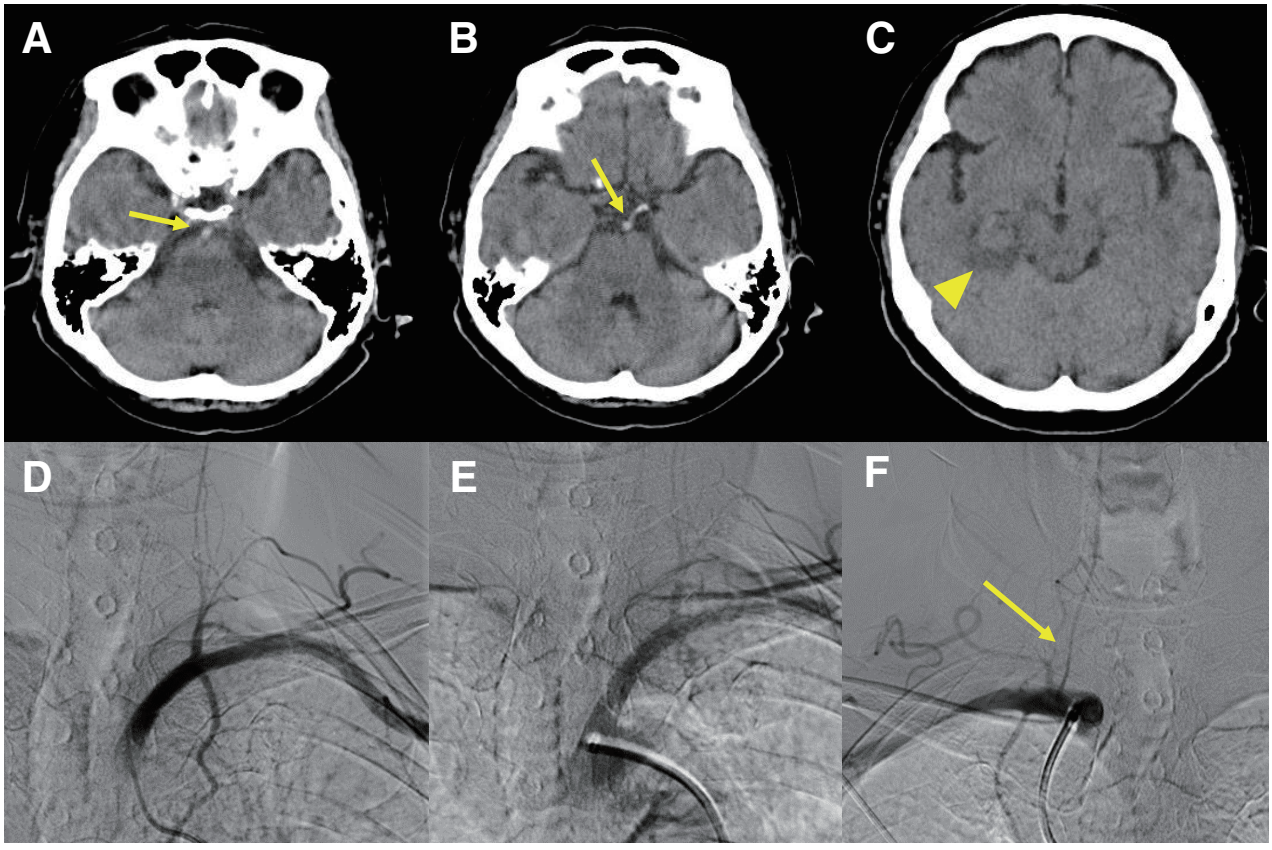

Fig. 1 (**A**–**C**) Preoperative head CT revealed early ischemic changes in the right parahippocampal gyrus (arrowhead) without intracranial hemorrhage and showed the hyperdense vessel sign in the BA and the left posterior cerebral artery (arrows). (**D** and **E**) Preoperative left subclavian artery angiography showed no evidence of the left VA. (**F**) Right subclavian artery angiography revealed the hypoplastic right VA (arrow). BA: basilar artery; VA: vertebral artery

of consciousness and was transferred to our hospital within 228 min from symptom onset. Physical examination revealed a National Institutes of Health Stroke Scale (NIHSS) score of 35 and Glasgow Coma Scale (GCS) score of 7 (E2V1M4), along with quadriparesis and anisocoria. Head CT revealed a low-density lesion in the right parahippocampal gyrus without intracranial hemorrhage or obvious early ischemic changes in the brainstem. Head CT also showed a hyperdense vessel sign in the BA, due to a thrombus approximately 30 mm in length from the mid BA to the P1 segment of the left posterior cerebral artery (**Fig. 1A**–**1C**).

The patient was diagnosed with acute BA occlusion due to cardiogenic cerebral embolism with atrial fibrillation and MT was performed; the time from symptom onset to groin puncture was 258 min. Intravenous tissue-plasminogen activator (t-PA) injection was not performed because of her advanced age and severe neurological symptoms. We planned to navigate a 6-Fr Flexor Shuttle guiding sheath 80 cm (Cook Medical, Bloomington, IN, USA) into the dominant left VA via the right femoral artery under local anesthesia. For this, we first performed left subclavian artery angiography, which showed no evidence of a left VA (**Fig. 1D** and **1E**). Next, we performed right subclavian artery angiography, which showed a hypoplastic right VA that was too small to allow navigation of a large bore aspiration catheter (**Fig. 1F**). The left VA was thought to directly originate from the aortic arch between the left CCA and left subclavian artery, although the origin of the left VA could not be navigated by the catheter. Finally, the catheter was navigated into the left CCA and angiography was performed, which showed the origin of the left VA from the PPA, which itself originated from the left ECA at the level of the C2 vertebral body (**Fig. 2A** and **2B**).

Subsequently, a 6-Fr Flexor Shuttle guiding sheath was easily inserted into the PPA. Since angiography showed occlusion of the BA immediately distal to the anterior inferior cerebellar artery (AICA) (**Fig. 2C** and **2D**), a Penumbra 5MAX ACE 60 (Penumbra, Alameda, CA, USA) aspiration catheter was navigated to the occlusion site with a coaxial system using a Velocity microcatheter (Penumbra Inc.) and Asahi Chikai micro guidewire (Asahi Chikai, Asahi Intecc, Aichi, Japan) (**Fig. 3A** and **3B**). After the 5MAX

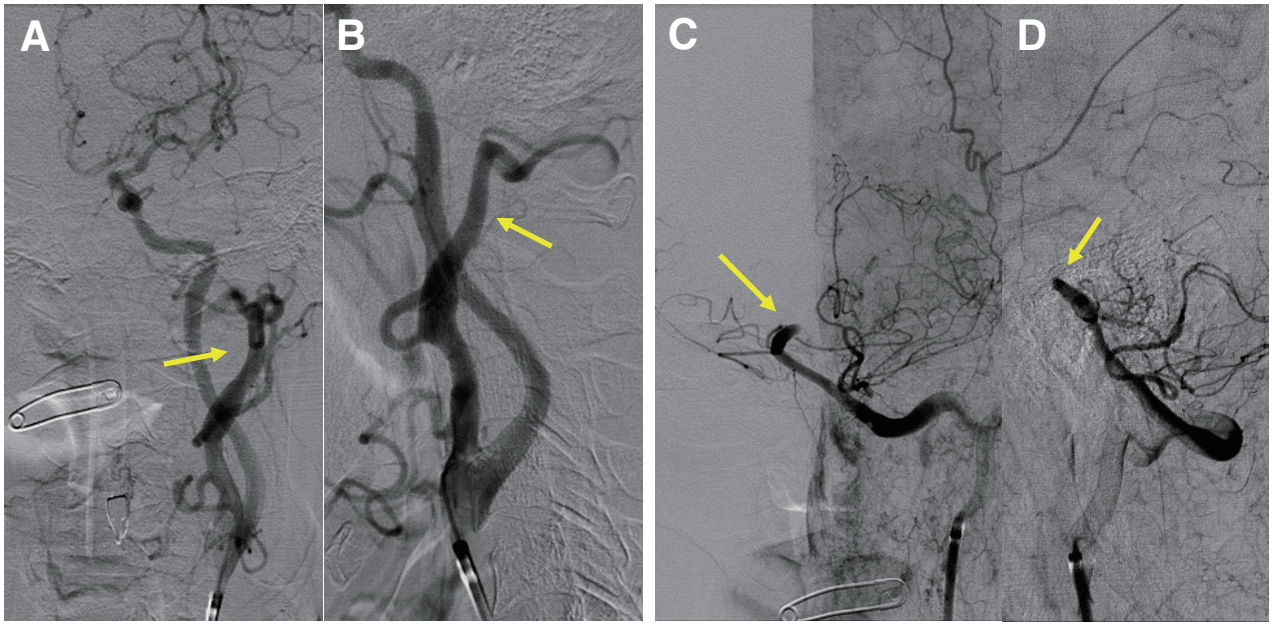

Fig. 2 Preoperative DSA of the left CCA. (**A**) AP view and (**B**) lateral view: showed the type 1 persistent PPA originating from the left ECA at the level of the C2 vertebral body that coursed between the C1 arch and occipital bone (arrows). Preoperative DSA of the PPA. (**C**) AP view and (**D**) lateral view: the BA was occluded just distal to the AICA (arrows). AICA: anterior inferior cerebellar artery; AP: anteroposterior; BA: basilar artery; CCA: common carotid artery; ECA: external carotid artery; PPA: proatlantal artery

ACE aspiration catheter contacted the clot, aspiration was commenced with a MAX pump (Penumbra Inc.) for 120 sec, and the aspiration catheter was withdrawn into the guiding sheath. A large red thrombus with a total length of 28 mm was retrieved (**Fig. 3C**) and successful recanalization of the BA was achieved at the first thrombectomy attempt, although with the subsequent development of distal occlusion of the branches of the posterior cerebral arteries (PCAs) bilaterally (Thrombolysis in Cerebral Infarction scale 2b) (**Fig. 3D** and **3E**). The time from groin puncture to recanalization was 27 min (time from puncture to the guide catheter reaching the PPA was 19 min), and head CT immediately after the procedure showed no sign of intracranial hemorrhage.

MRA performed the next day showed good recanalization of the BA and absence of bilateral posterior communicating arteries (PCoAs) (**Fig. 3F**), although head MRI FLAIR images revealed a large hemorrhagic infarction in the pons (European Cooperative Acute Stroke Study [ECASS] criteria[6]: PH2) and infarction in the right temporal and bilateral occipital lobes (**Fig. 3G** and **3H**). Cervical MRA showed that the left PPA originated from the ECA at the level of the C2 vertebral body (**Fig. 4A**), ascended to the suboccipital space without passing through the transverse foramen of the atlas (**Fig. 4B**) and coursed between the atlas and occipital bone to enter the foramen magnum (**Fig. 4C**). The patient's symptoms did not improve despite successful recanalization. She was transferred to a long-term hospital on the 37th day after symptom onset, and her modified Rankin Scale (mRS) score was five at 90 days after the symptom onset.

Discussion

Primitive CVAs are formed in the early embryonic period and usually regress during subsequent embryonic development.[7] The four primitive CVAs occasionally persist into adulthood,[2] forming the persistent trigeminal artery (0.5%–0.7%),[2] persistent otic artery, persistent hypoglossal artery (0.027%–0.29%),[2] and PPA (0.02%).[1] PPAs are classified into two types. A type 1 PPA arises from the ICA, ECA, or CCA and ascends to the level of the suboccipital space without passing through the transverse foramen of the atlas. The artery courses between the C1 arch and occipital bone, meets the extracranial VA, and enters the foramen magnum. On the other hand, a type 2 PPA originates from the ECA, passes through the C1-2 interspace and the transverse foramen of the atlas, and joins the extracranial VA before entering the foramen magnum.[2,8] In this case, since the PPA ascended to the suboccipital space without passing through the transverse foramen of the atlas and coursed between the atlas and occipital bone to enter the foramen magnum (**Fig. 4**), it was diagnosed as a type 1 PPA.

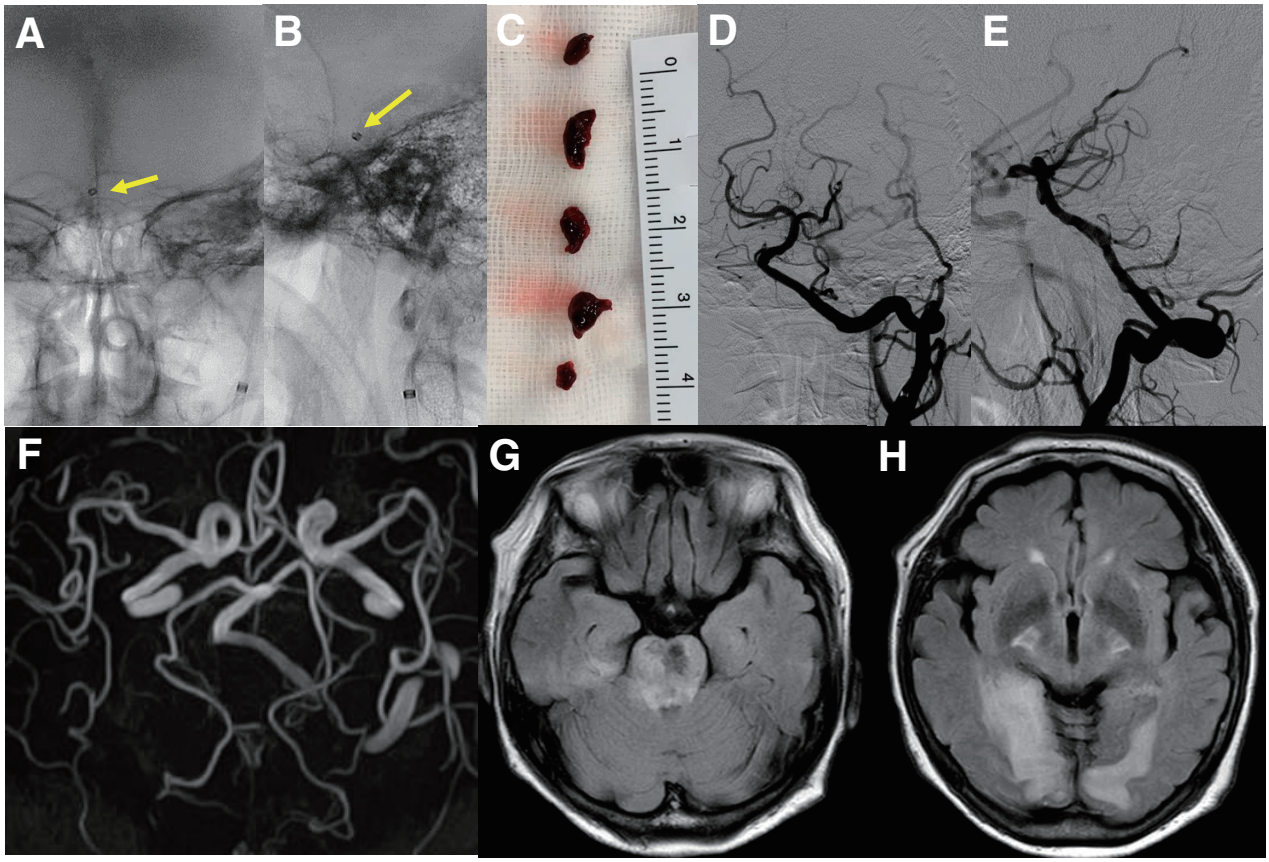

Fig. 3 (**A** and **B**) Digital angiogram during the procedure (**A**: AP view and **B**: lateral view): the Penumbra 5MAX ACE 60 catheter was navigated into the occluded BA (arrow). (**C**) A red thrombus 28 mm in length was retrieved by MT. (**D** and **E**) DSA of the left ECA immediately after the first attempt (**D**: AP view and **E**: lateral view): recanalization of the BA was achieved successfully, although with distal occlusion of the posterior cerebral arteries bilaterally. (**F**) MRA performed the day after thrombectomy showed good recanalization of the BA and absence of bilateral PCoAs. (**G** and **H**) MRI FLAIR images on the day after thrombectomy revealed a large hemorrhagic infarction in the pons (ECASS criteria: PH2) and infarction in the right temporal and bilateral occipital lobes. AP: anteroposterior; BA: basilar artery; ECA: external carotid artery; ECASS: European Cooperative Acute Stroke Study; MT: mechanical thrombectomy; PCoAs: posterior communicating arteries

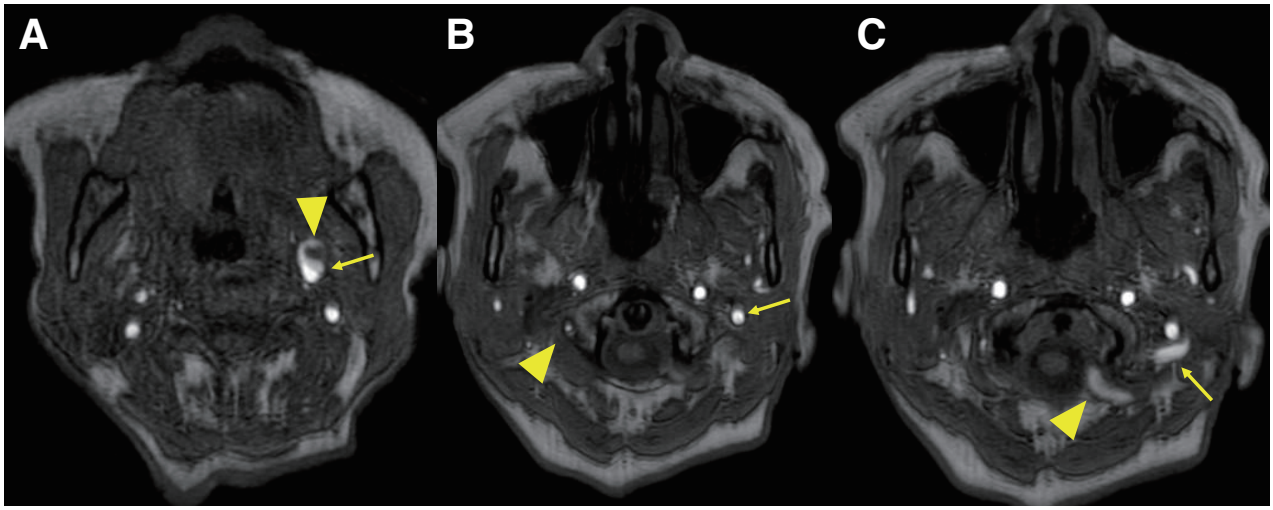

Fig. 4 Cervical MRA source image on the day after thrombectomy. (**A**) At the level of the C2 vertebral body: the left PPA (arrow) originated from the left ECA (arrowhead). (**B**) At the level of the C1 vertebral body: unlike the contralateral VA (arrowhead), the left PPA (arrow) ascended without passing through the transverse foramen of the atlas. (**C**) At the level of the foramen magnum: the PPA (arrow) coursed between the C1 arch and occipital bone to enter the foramen magnum (arrowhead). ECA: external carotid artery; PPA: proatlantal artery; VA: vertebral artery

Although many cases of PPA have been found incidentally, there are only a few reports of cases in which the PPA was directly related to the disease. Several PPA cases with carotid artery stenosis,[9] VA stenosis,[10] and vertebrobasilar aneurysm[11] have been successfully treated using endovascular methods. On the other hand, cases such as ours, with acute BA occlusion and a PPA, are extremely rare and only three such cases have been reported to date.[3–5] In 1988, Sato et al. reported a case of BA occlusion with a right type 2 PPA that resulted in infarction of the cerebellum and pons.[3] In 1993, Bahşi et al. also reported a case of "top of the Basilar syndrome" with a left type 1 PPA that led to infarction of the medial basal thalamus bilaterally.[4] In both cases, thrombolysis or MT was not performed and the patient's severe disability persisted. Zhao et al. were the first to report a case of acute basilar occlusion with type 1 PPA for which stent retriever thrombectomy was performed.[5] In their case, although the BA was completely recanalized, the patient died due to hemorrhagic brainstem infarction, suggesting that poor collateral circulation because of the absence of bilateral PCoAs might have resulted in fatal brainstem infarction.

Recently, the results of two randomized controlled trials showing the efficacy of MT in patients with BA occlusion have been published: in the ATTENTION trial[12] conducted in China, patients within 12 hours from onset were randomized in a 2:1 ratio to receive endovascular or medical therapy, and the rate of patients with good functional status (mRS score at 90 days of 3 or lower) was significantly higher in the endovascular group (46% vs. 23%, adjusted risk ratio 2.06, 95% confidence interval [CI] 1.06). The mortality rate was also significantly lower in the endovascular group (37% vs. 55%, adjusted risk ratio 0.66, 95% CI 0.52–0.82). In the BAOCHE trial as well,[13] which examined 217 patients within 6–24 hours of disease onset, the percentage of patients with good functional status was significantly higher in the endovascular treatment group (46% vs. 24%, adjusted risk ratio 1.81, 95% CI 1.26–2.60).

In our case, although successful recanalization was achieved, the patient's neurological symptoms did not improve due to several possible reasons. Good collateral circulation and distal BA occlusion are known to be independent predictors of clinical outcomes in patients with acute BA occlusion.[14] In this case, the BA was occluded immediately distal at the origin of the AICA, suggesting the possible presence of a long clot along the entire length of the BA, occluding the perforators from the BA and resulting in the large brainstem infarction. Second, undeveloped bilateral PCoAs resulted in a poor collateral circulation (**Fig. 3F**). Typically, the primitive CVAs rapidly regress as the PCoAs and vertebrobasilar arteries develop,[8] and PPA is often accompanied by PCoAs hypoplasia.[5] Finally, it took a long time to identify the origin of the left VA during the procedure since no head or cervical angiography was performed preoperatively. Yuan reviewed 980 cases with a single aberrant origin of left VA, among which aortic arch branching of the VA was identified in 955 (97.4%) cases.[15] On the other hand, a left ECA origin of the VA and a left CCA origin were reported in two cases (0.2%) each. This suggests that in cases in which the left VA does not originate from the left subclavian artery or aortic arch, it might be possible to identify the left VA using a left common arteriogram.

Conclusion

In this case, although MT achieved successful recanalization of the BA via the PPA, the patient's clinical symptoms did not improve, probably because of poor collateral circulation due to undeveloped PCoAs bilaterally or due to the long length of the occluded segment in the BA due to the large thrombus. In patients with acute vertebro-BA occlusion, if the VA does not originate from the subclavian artery or aortic arch, the presence of a primitive CVA should be considered.

Disclosure Statement

None of the authors have any conflicts of interest in relation to this article.

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