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Persistent trigeminal artery as a rare cause of vertebrobasilar insufficiency

K. Sulima et al., Persistent trigeminal artery

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ABSTRACT

The persistent trigeminal artery (PTA) is the most common fetal carotid- basilar anastomosis which may persist into adult life. In the literature there are numerous papers referring to the anatomical characteristics of this anomalous vessel. In the majority of cases PTA is an incidental finding and its clinical significance has been debated. Some authors describe the coincidence of PTA with various clinical syndromes. However, there are few reports linking PTA with the symptoms of vertebrobasilar insufficiency. We present a patient with a 3-year history of recurrent dizziness and impaired vision, precipitated by physical activity, in whom neuroimaging techniques revealed a persistent left trigeminal artery (PTA). In our case the occurrence of PTA might have been related to the clinical manifestation and the potential mechanism will be discussed.

Key words: persistent trigeminal artery, computed tomography angiogram, stroke, magnetic resonance imaging, vertebrobasilar insufficiency

INTRODUCTION

The persistent trigeminal artery (PTA) is the most common remnant of the primitive circulatory system that unites the internal carotid (anterior) and vertebrobasilar (posterior) systems. The PTA contributes to about 85% of known primitive persistent anastomoses [1,25]. The reported incidence of the PTA ranges from 0.1 to 0.6% [7,24]. Most cases are detected incidentally during brain imaging performed for unrelated reasons, so the real frequency can

be higher, based on unrecognized cases. The PTA is usually asymptomatic, however in some cases may be connected with III or VI nerves palsies, trigeminal neuralgia, vertigo, ataxia, stroke, migraine headache and other neurological symptoms [9,14,30,35]. It may be also associated with other vascular abnormalities such as basilar hypoplasia or anatomical variations of aortic arch and its branches [10,29].

CASE REPORT

A 33-year-old man, with no prior comorbidities, presented to our clinic with a 3-year history of recurrent dizziness and impaired vision. The symptoms were transient, lasting usually over a few seconds, triggered by moderate physical activity, especially walking. Head rotation did not aggravate the symptoms. There was no prior history of arm claudication, as well as loss of consciousness or seizures. The patient was an underground copper miner, potentially exposed to dust and toxins. No neurological deficit was found on examination. Magnetic resonance imaging (MRI) of the brain, with and without contrast enhancement, was within the normal limits. A computed tomography angiogram (CTA) was performed, revealing a persistent left trigeminal artery (PTA), an anomalous vessel extending from the internal carotid artery (ICA) to the basilar artery (BA) (Fig.1-5). The PTA arose from the junction between petrous and cavernous ICA, and ran posterolaterally along the trigeminal nerve, its diameter was about 2-3 mm. The PTA filled superior cerebellar arteries (SCAs), while the patent left posterior communicating artery (PCoA) supplied the posterior cerebral arteries (PCAs). According to its course and connections, the vessel was classified as Saltzman type II. The examination also showed the focal stenosis of the right vertebral artery (VA) at the C4 level, the absence of the right posterior communicating artery (PCoA) and hypoplasia of A1 segment of anterior cerebral artery (ACA) (Fig. 2). The scheme 1 is a pictorial presentation of the cerebral vasculature anatomy in our patient. No other variations were found in the vessels supplying the central nervous system. The patient was treated conservatively with acetylsalicylic acid (75 mg per day) and cinnarizine with dimenhydrinate (20+40 mg three times per day) as a short- term therapy for dizziness. The potential benefits of surgical procedure would not justify the possible risk of such treatment.

DISCUSSION

There are many reports regarding intracranial vascular development, and one of the most significant was made by Padget in 1948 [2,32]. During the evolution of the vertebrobasilar arterial system, the hindbrain is supplied predominantly by four fetal carotid-

basilar anastomoses: the trigeminal, otic, hypoglossal, and proatlantal intersegmental arteries, arising at the 3 mm embryonic stage. The above-mentioned connections regress with the subsequent development of posterior communicating arteries and basilar artery and should disappear by the 14 mm stage [2]. Failure of this process results in persistence of these vessels into adult life. Within four persistent fetal carotid- basilar anastomoses, the trigeminal artery is the most common.

The PTA was firstly reported by Richard Quinn in 1844 during an autopsy [35]. In 1950 the PTA was recognized by Sutton, based on angiography [35].

The PTA typically arises from the posterior or lateral part of intracavernous (C4) segment of the internal carotid artery (ICA) and terminates at the basilar artery, between the first part of the SCA and anterior inferior cerebellar artery (AICA) [4,12,34]. In some cases pontine perforating branches of PTA are observed [3,23,33]. The incidence of PTA varies from 0.1-0.6% [11] and according to some studies is higher in women [5,22]. In most cases the right or none- sided predominance of the PTA is detected.

The most common classification system, based on angiographic characteristics, was introduced by Georg-Frederik Saltzman in 1959 [4]. It divides the PTA into three groups according to its relationship to the basilar artery. In Saltzman type I, the PTA enters the basilar artery between the superior cerebellar artery (SCA) and the inferior anterior cerebellar artery (AICA), supplying both posterior cerebral (PCA) and superior cerebellar arteries (SCA). The basilar artery (BA) is typically hypoplastic and the ipsilateral posterior communicating artery is absent. In Saltzman type II, the PTA provides the anterior superior cerebellar artery (ASCA), whereas the posterior cerebral arteries arise from the posterior communicating arteries. The first segment of BA is missing. Saltzman type III, the rarest type, is considered to be a combination of types I and II. In type III PCA is supplied by the ipsilateral posterior communicating artery (PCoA), the other PCA and both SCA being supplied by the PTA. Saltzman type I is the most prevalent and accounts for about 24% of all cases [29,35].

In 1998 Salas proposed a different classification, dividing the PTA into two types based on its relationship to the abducens nerve: the lateral (petrosal) and medial (sphenoidal). The lateral type is recognized when the artery courses laterally to the abducens nerve and pierces the dura mater just medially to the sensory root of the trigeminal nerve. The second, medial type, is recognized when the artery courses medially to the abducens nerve, penetrates the dorsum sellae and joins the distal third of the basilar artery [33]. The study indicated that the latter type is more common [31,36].

Most cases of PTA are recognized incidentally in imaging diagnostic tests performed for other reasons. Typically PTA is associated with BA hypoplasia, the observation being confirmed in the large neuroimaging series [10,31]. There are some reports in the literature regarding the correlation of the PTA with other abnormalities in the circle of Willis, such as the absence of the ipsilateral posterior communicating artery, vertebral artery or basilar artery. Due to embryonic error in vasculogenesis, the PTA is frequently observed in PHACE syndrome [19]. The connection with the increased risk of aneurysm is controversial. The high incidence of aneurysm is explained by hemodynamic changes in blood flow, which promote aneurysm formation [31]. In an older study [1,18], the prevalence of associated intracranial aneurysms in patients with PTA was reported to be up to 14-32%. In the newest reports based on MRI examination, the prevalence is estimated to be approximately 16% [12]. However, Meckel et al. suggested that the real frequency of aneurysm coexisting with the PTA does not differ from that in the general population (4.2% versus 3.7%) [25,29]. Aneurysm connected with the PTA has a lot of potential sites. Behari described an aneurysm on the posterior communicating artery, Maeshima showed a case of PTA connected with multiple aneurysms on ACA and MCA [8,28]. Bechri in 2020 reported a case of a posterior meningeal artery aneurysm coexisting with PTA [7]. In a great number of cases the persistent trigeminal artery is an incidental discovery and its clinical significance has been debated. The symptoms may depend on associated vascular abnormalities or their localization. The PTA has been identified as a rare cause of cranial nerves dysfunction, including incomplete oculomotor and abducens nerve palsies, trigeminal neuralgia and hemifacial spasm. The ischemic events related to the PTA are rare and can occur through the steal phenomenon or thrombosis in anterior circulation [16,21]. Typically, persistent trigeminal artery aneurysms (PTAA) are discovered incidentally in patients with subarachnoid hemorrhage (SAH) due to the rupture of a simultaneous aneurysm in another localization. Ruptured PTAA can be presented with symptoms of headache and posterior cranial fossa symptoms, according to the SAH [29]. In the case of large and giant cavernous PTAA, cranial neuropathy or facial pain can be observed [13]. Furthermore, PTA was postulated as a potential cause of vertebrobasilar insufficiency (VBI), which could be defined as inadequate blood flow through the posterior circulation system, without actual infarction, with a temporary inability to meet cerebral metabolic needs as a consequence [20]. Posterior circulation supplies the brainstem, cerebellum, thalamus, occipital lobe, some portions of temporooccipital and parietooccipital junctions and even the inner ear through the labyrinthine artery [6]. Dizziness is the most common symptom of VBI, but the diversity of brain regions being supplied by vertebrobasilar

vasculature explains the variety of complaints, such as diplopia, blindness, visual field deficits, imbalance, and ataxia [20]. Patients with VBI usually present more than one symptom at the same time. The most frequent pathology underlying VBI is atherosclerosis, however other causes such as embolism, arterial dissection, migraine and fibromuscular dysplasia may occur [27]. As during embryonic development PTA originates from ICA, the physiological direction of blood flow through PTA is mostly from the internal carotid artery (ICA) to the basilar artery (BA) [6,15]. Nevertheless, the reverse of this direction may occur in certain clinical situations. In case of severe stenosis or occlusion of the ipsilateral internal carotid artery, the reversal of flow through PTA may play a protective role and be sufficient to perfuse ICA and its branches, keeping the patient free of hypoperfusion symptoms [26]. Conversely, if PTA coexists with other anomalies of the intracranial vessels, such as hypoplastic vertebral artery, ICA may be the sole supply to the posterior circulation system. In that case if carotid stenosis occurs, the patient may develop symptoms of vertebrobasilar insufficiency [6]. However, our patient did not have any risk factors for cardiovascular disease, and there were also no radiologic signs of atherosclerosis. The review of the literature revealed only one previously reported case linking PTA with VBI symptoms, without the simultaneous presence of carotid pathology, in which dilatation of PTA and basilar artery (BA) was the speculated mechanism [17]. In our patient the lack of the right posterior communicating artery (PCoA), together with the stenosis of the right vertebral artery (VA), might have resulted in the left ICA being the main supply to the posterior circulation system. In the case of the exercise- induced increase of metabolism in the anterior portion of the brain, PTA might have been insufficient to perfuse the posterior fossa region of the brain, leading to vertebrobasilar insufficiency symptoms. We could not exclude the potential reverse of blood flow direction in this case, from the vertebrobasilar system to the carotid artery.

CONCLUSIONS

Due to the high prevalence of dizziness in the general population, awareness of its rare causes seems to be crucial. The persistent trigeminal artery (PTA) may play a role in the production of vertebrobasilar insufficiency (VBI) symptoms. In patients with VBI of unknown origin, a computed tomography angiogram (CTA) should be considered as a diagnostic procedure.

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Figure 1. CTA axial view show persistent trigeminal artery (PTA) on the left (arrow). It is the most common variant of Persistent carotid basilar anastomoses (PCBA).

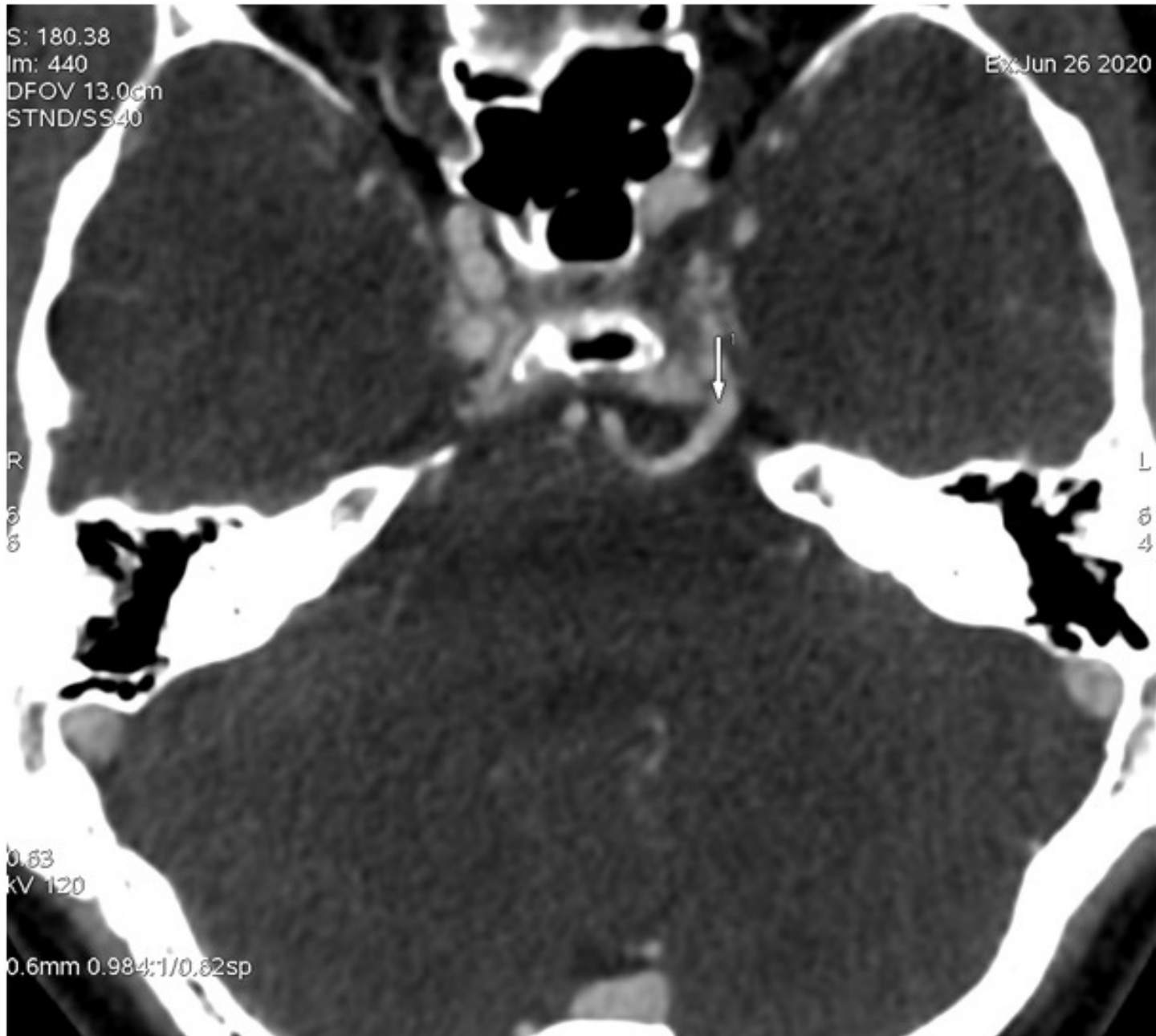
Figure 2. MR. Axial T 2-weighted scan. Persistent trigeminal artery (PTA) on the left (arrow).

Figure 3. CTA, MIP reconstruction. PTA fills superior cerebellar arteries, PCAs supplied via patent PCoA on the left (star). The absence of right PCoA (arrow).

Figure 4. Volume rendering 3D reconstruction, cerebral angiogram based on CTA examination. Bigger arrow PTA (connection BA-ICA), smaller arrow PCoA (connection PCA-ICA), both on left side.

Figure 5. Posterior circulation CTA, MIP oblique reconstruction.

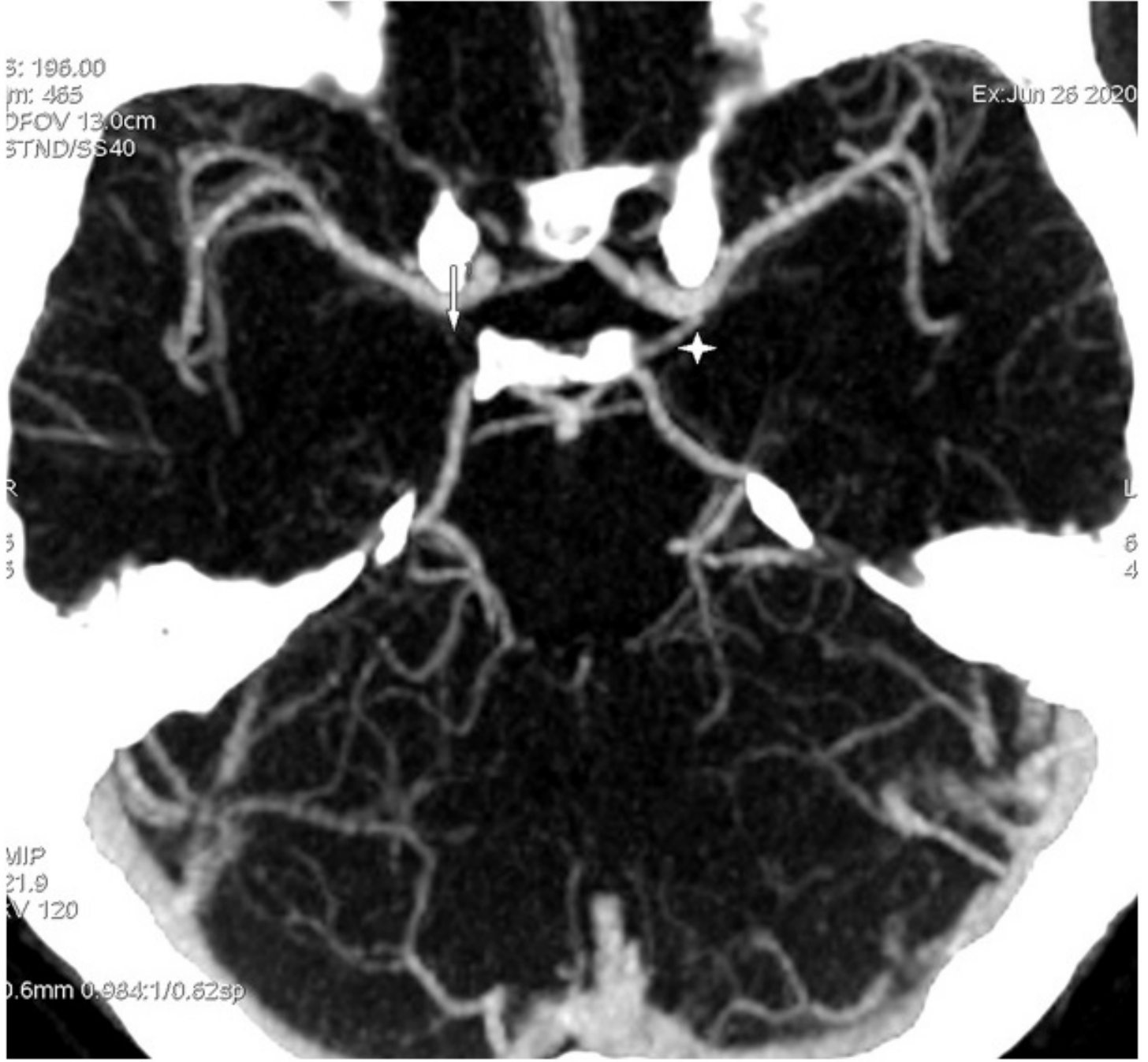
Scheme 1. Schematic presentation of the cerebral vasculature anatomy in our patient. ACA A1: A1 segment of anterior cerebral artery; ACA A2: A2 segment of anterior cerebral artery; ACoA: anterior communicating artery; AICA: anterior inferior cerebellar artery; BA:basilar artery; MCA: middle cerebral artery; PCA: posterior communicating artery; PTA: persistent trigeminal artery; SCA: superior cerebellar artery ;VA: vertebral artery





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DFOV 13.0cm
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Ex: Jun 26 2020

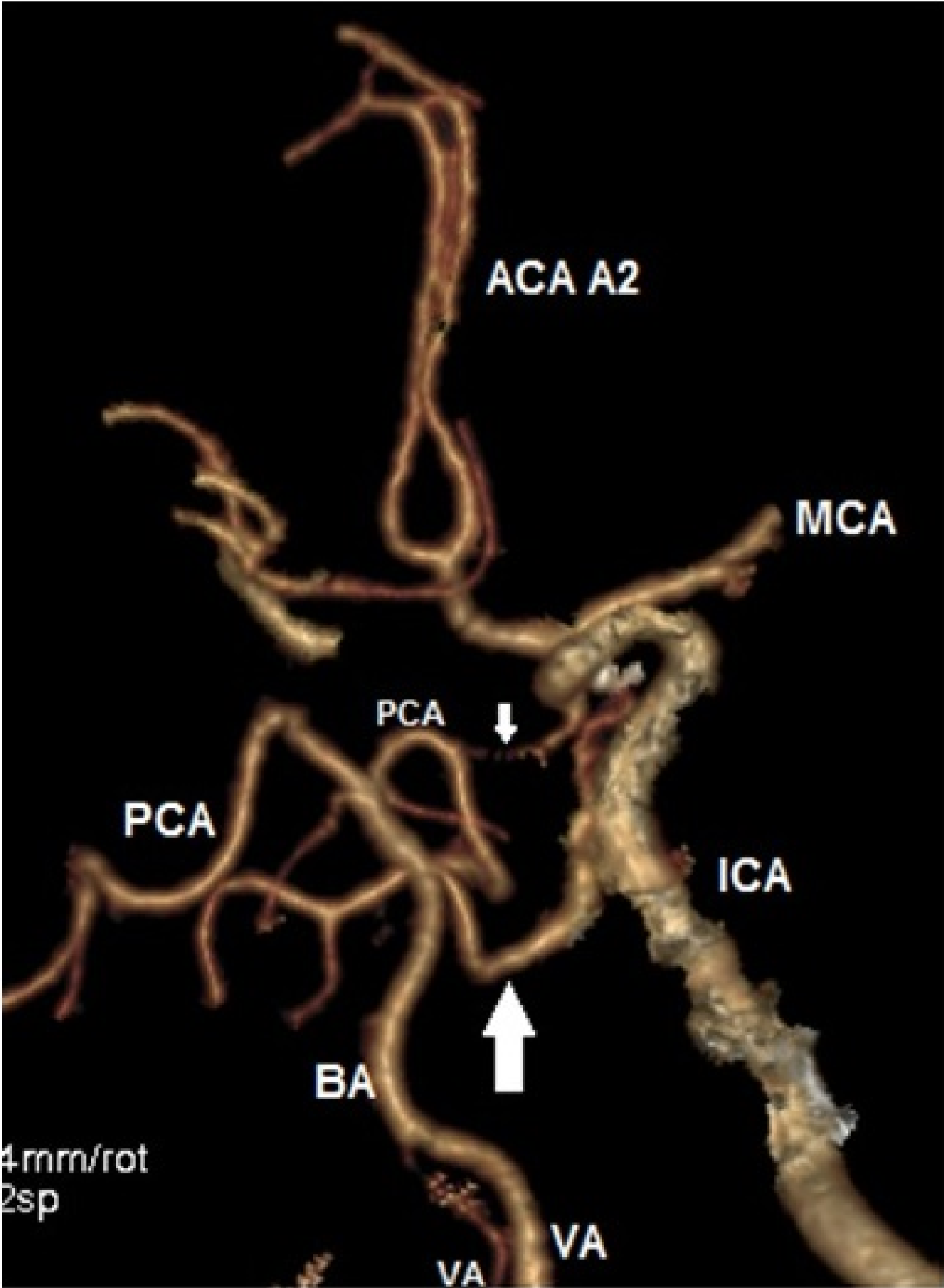


MIP
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R
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3

L
6
4



74 (col)
14.8cm
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