

Posterior Circulation Ischemic Stroke in Association with Vertebral Artery Duplication: A Case Report and Review of Literature

Sir,

Duplication of the vertebral artery (VA) refers to a condition where VA has a dual origin with variable course and different levels of fusion at cervical vertebrae. Duplication of the VA is clinically asymptomatic in most cases. It is usually detected as an incidental finding in autopsy series, angiographic studies, computed tomography (CT) angiography, and magnetic resonance angiography. Vertebral artery duplication may be associated with an increased risk of thrombosis or dissection due to minor neck trauma, leading to vertebrobasilar insufficiency presenting with neurological symptoms such as dizziness, vertigo, headache, or even posterior circulation stroke. To our knowledge, only a few cases of vertebral artery duplication associated with posterior circulation stroke are reported and we could not find any such case report published from India, so possibly, our case is the first.

A 37-year-old male patient with no vascular risk factors presented to the emergency department with a sudden onset of vision loss in the right hemifield, which he noticed while driving back home from work for the last one day. He had experienced a similar episode associated with numbness in the right half of his body, including face, the day before vision loss, which had resolved completely over a period of 12 h. There was no history of dizziness or vertigo, gait imbalance, weakness in any limb, or altered mental status. The examination revealed normal higher mental functions except for impaired recent memory and right

homonymous hemianopia; the rest of the examination was normal. A magnetic resonance imaging (MRI) brain diffusion weighted image with apparent diffusion coefficient (DWI-ADC) image [Figure 1a] showed restricted diffusion in the left medial temporal lobe and left occipital lobe, suggestive of posterior cerebral artery (PCA) territory infarcts likely to be embolic, as the infarct location was cortical and involved distant branches of the PCA. A CT angiogram (CTA) of the brain, neck, and arch of the aorta showed a small caliber lateral limb of the left vertebral artery (VA) originating from the left subclavian artery and a larger caliber medial limb of VA, which was discontinued with a distal segment and a proximal stump at the arch of the aorta proximal to the origin of the left subclavian artery [Figure 1b]. Sagittal and axial images showed the small caliber lateral limb of VA originating from the left subclavian entering at the C6 transverse foramen, merging with the distal end of the medial limb at C5-6 level and continuing as the V2 segment of the left VA, suggesting a duplicated VA [Figure 1c and d]. As the VA originating from the arch showed discontinuity (stump at the arch and a distal segment near the merging point), dissection of this segment as the etiology of the stroke was thought of. Digital subtraction angiography (DSA) was performed, and it confirmed the CTA findings with tapered obstruction of VA proximal to the merging point [Figure 1e]. The patient reported a history of frequently falling asleep in his chair while working at a surveillance station, with his neck extended over the edge

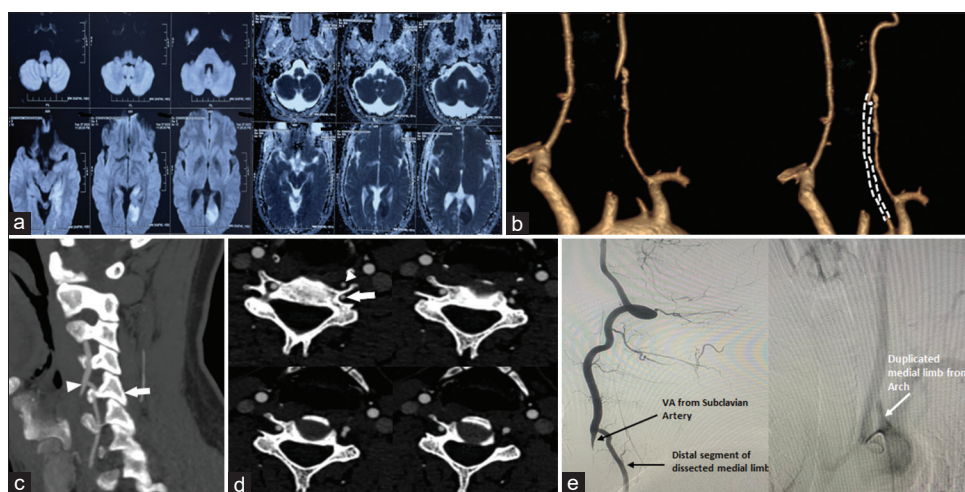


Figure 1: (a) MRI brain showing restricted diffusion in left PCA territory involving left medial temporal lobe and occipital lobe. (b) 3D reconstructed image of CT angiogram of neck showing the dissected medial limb and its probable course (dotted lines). (c) CT angiogram serial axial view shows medial limb (arrowhead) and lateral limb inside the transverse foramen (arrow) uniting at C5-6 level. (d) CT angiogram sagittal view showing the two limbs of duplicated VA uniting (arrowhead) after the lateral limb enters at C6 transverse foramen (arrow). (e) DSA confirming the CTA findings of distal tapering end and proximal origin from arch of medial limb of duplicated vertebral artery and lateral limb arising from subclavian artery

of the chair during night shift. The patient was started on dual anti-platelet therapy, and there were no further episodes of TIA or exacerbation of symptoms during the hospital stay.

The VA is formed by fusion of the longitudinal anastomoses of the cervical intersegmental arteries, which branch off from the primitive paired dorsal aorta during embryological development. In normal situations, almost all intersegmental arteries regress, except for the seventh intersegmental artery (i.e. the sixth cervical intersegmental artery), which forms the proximal portion of the subclavian artery.^[1] Of the several mechanisms that may give rise to a separate origin of the VA, one explanation suggests that the primitive dorsal aorta does not regress with the two intersegmental arteries connecting to the VA, giving rise to duplication of the VA. Another possibility is a failure of the regression of the fourth, fifth, or sixth intersegmental arteries (i.e. the third, fourth, or fifth cervical intersegmental arteries)^[2] resulting in an additional origin of the VA along with a normal seventh intersegmental artery. Persistence of the left fourth or fifth (or less frequently third) cervical intersegmental artery may result in an aortic origin or dual origin of the left VA. During duplication of the VA, one limb can originate from the subclavian artery, whereas the second limb can originate from the aortic arch, subclavian artery, thyrocervical trunk, or innominate trunk.^[2] Rarely, both limbs of a duplicated left VA originate from the aorta. Usually, the medial limb enters the higher vertebral transverse foramen, which is consistent with the theory of intersegmental vessel regression failure, and results from intersegmental arteries that accompany the cervical nerve roots.

Table 1 reviews the case reports of duplicated VA associated with posterior circulation stroke due to dissection or thrombosis of duplicated VA. Melki *et al.*^[3] and Meila *et al.*^[4] explained the pathophysiologic mechanism of a duplicated artery

having vascular dysgenesis with histological defects during embryologic development or a weaker vessel wall due to abnormal hemodynamics, leading to possible dissection. In our case too, we suspect a similar pathophysiologic mechanism of dissection of duplicated VA medial limb due to trivial trauma of repeated neck hyperextension by virtue of his occupation presenting as thromboembolic posterior circulation stroke. Unlike previous cases, our patient had a medial limb of duplicated left VA arising from the aortic arch posterolateral to the origin of the left common carotid artery and a lateral limb arising from the left subclavian artery. Both limbs of duplicated VA were merging at a lower cervical vertebral level (C6) compared to previous cases. In a duplicated VA, the other limb may be collateral to the distal VA when the proximal segment of the subclavian artery or the VA originating from the subclavian artery exhibits stenosis.^[5] Thrombosis or dissection of one limb of a duplicated VA may result in less severe symptoms compared with other types of VA thrombosis or dissection because of a collateral pathway via another limb of the duplicated VA. Operative correction of a kinked duplicated origin of the VA, which was suspected to underlie disabling dizziness in a 70-year-old patient, was performed despite the absence of any other evidence of its benefits.^[6] Polguy *et al.*^[7] reported that the lumen of the duplicated VA is smaller than normal; hence, this variant has clinical implications and should be considered when a VA needs catheterization. In such patients, interventional procedures should be performed from the normal side, if possible. Satti *et al.*^[8] suggested that VA can be easily damaged during severe cervical spine injuries with rapid sublaxation, deceleration, fracture through the transverse foramen, or cervical spine flexion. The additional point of attachment of the VA predisposes individuals with a duplicated origin of the VA to experience more severe outcomes from the kind of contusion that most commonly occurs during motor vehicle accidents.

Table 1: Reported cases of duplicated vertebral artery presenting with posterior circulation stroke

Case	Age	Sex	Clinical presentation	Precipitating factor	Ct/mri	Cause	Side of dupli-cated VA	Site of origin of duplicated limb	Level of merging of duplicated VA limbs
Dare <i>et al.</i> ^[1]	38	M	Loss of consciousness	Sexual intercourse	Hypo-attenuation in basis pontis	Rt VA dissection distal to merging	Right	Both limbs from subclavian artery	C4
Melki <i>et al.</i> ^[3]	51	M	Acute vertigo with transient cervicgia	Minor neck trauma	Cerebellar vermis infarct	Rt VA medial limb dissection	Right	Both limbs from subclavian artery	Not reported
Mahmutyazicioğlu <i>et al.</i> ^[9]	62	M	Recurrent episodes of vertigo, nausea, weakness	Paroxysmal atrial tachycardia	Not reported	Thrombosis of VA	Left	Not reported	C2
Kissel <i>et al.</i> ^[10]	56	F	Right hemiparesis and diplopia	Focal stenosis in close contact with the hyoid bone	Left paramedian midbrain infarct	Rt VA duplicated segment dissection	Right	Not reported	Not reported
Our case	37	M	Right homonymous hemianopia	Repeated neck hyperextension	Left occipito-temporal infarct	Left VA duplicated limb dissection	Left	Medial limb-aortic arch, lateral limb-left subclavian artery	C6

Duplication of VA is asymptomatic in most instances and diagnosed incidentally; however, knowledge of this variant is important as in rare cases, vertebral artery duplication may be associated with an increased risk of vertebrobasilar insufficiency manifesting with diverse neurological symptoms including ischemic stroke and avoiding misinterpretation or injury of a duplicated VA during vascular head and neck surgery, spinal surgery, or cerebral angiography.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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