

# Unilateral Vertebral Artery Hypoplasia Masquerading as Benign Paroxysmal Positional Vertigo: A Case Report

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## ABSTRACT

Unilateral vertebral artery hypoplasia has been reported as possibly contributing to acute ischemia stroke or migraine with aura and could be related to episodic positional vertigo. An 88-year-old man presented with episodic vertigo whenever he rotated his head 90° to the right. He had experienced this for the last two years. Benign paroxysmal positional vertigo was initially suspected, but Epley's canalith repositioning procedures failed to cure the symptom. The symptom was finally confirmed as related to right vertebral artery hypoplasia (VAH) after he had received a battery of studies, including an electronystagmogram, a color-coded duplex sonogram, and a magnetic resonance imaging/ angiogram. Thus, conservative treatment and life-style change were recommended and led to a decrease in recurrence. Herein we report this case and the oto-neurological significance of vertebral artery hypoplasia. In conclusion, one-sided head rotation related to episodic vertigo might be attributable to VAH.

**Keywords:** Color-coded duplex sonogram, vertigo, flocculus, Epley's canalith repositioning procedure

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**E**pisodic vertigo might be attributed to cervical origins, peripheral vertigo, migrainous vertigo, vertebrobasilar artery insufficiency, dysautonomia causing orthostatic hypotension, intracranial hypotension, intracranial hypertension and other causes. In order to exclude all possible etiologies of episodic vertigo, further investigations include flexion and extension cervical radiographs, an electrocardiogram, a 24 hours' Holter, an electronystagmogram, a caloric test, a color-coded duplex sonogram, and an angiogram are required. In Taiwan, these studies are arranged in this order according to convenience and cost.

Vertebral artery hypoplasia (VAH), is defined as when the vertebral artery (VA) is less than 2.0 mm in diameter by time-of-flight magnetic resonance angiogram (TOF MRA), which has been reported to range 2-6% based on autopsy findings and angiograms. VAH has been reported as possibly contributing to acute ischemia stroke or migraine with aura.<sup>1,2</sup> This article

describes the importance of understanding the possible relationship between VAH and episodic positional vertigo and how this affects the approach to its treatment.

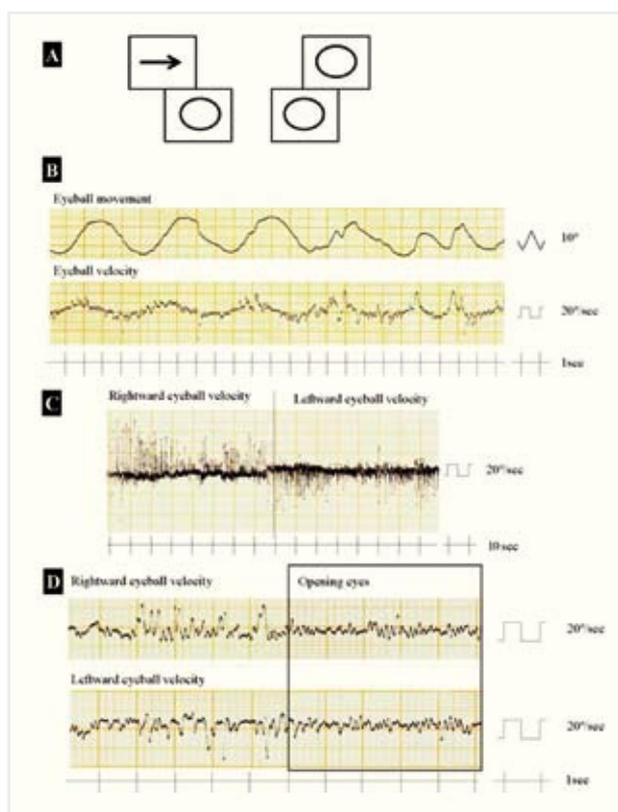
## CASE REPORT

An 88-year-old man, height 157 cm, weight 65 kg, and body mass index 26.4 kg/m<sup>2</sup> regularly took 25 mg of carvedilol for hypertension for four years. He was not addicted to cigarettes, alcohol, areca (betel nut) or other medication. However, he had had frequent episodic vertigo since April 2008. Simple vertigo, rather than any other neurologic signs, occurred 3-5 times per day when he rotated his head up to 90° to the right. The symptomatic duration was less than one minute. An otorhinolaryngologist initially suspected benign paroxysmal positional vertigo, but frequent Epley's canalith repositioning procedures did not cure his symptom. He took diphenidol (Cephadol 25 mg) and nicametate (Euclidan 50 mg) every day, but the symptom persisted. Nine months later, he visited our hospital.

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His blood pressure was 125/87 mmHg with a heart rate of 53/min. The otorhinolaryngologic examination did not show any abnormalities. There was no evidence of gaze nystagmus, but the rightward Dix-Hallpike positioning test showed leftward horizontal nystagmus (Fig 1A). The orthostatic hypotension test was negative. However, tests for hyperventilation and a squat to standing position also induced the leftward horizontal nystagmus. Flexion and extension cervical radiographs showed small osteophytes in all cervical joints. An electrocardiogram and 24 hours' Holter did not show any abnormality. An electronystagmogram revealed that pursuit (Fig 1B) and optokinetic nystagmus (Fig 1C) were abnormal. A caloric test (20°C tap water, 20 secs) showed bilaterally negative visual suppression (Fig 1D).

Color-coded duplex sonography showed atherosclerosis over all of the carotid arteries. The right VA was not measurable. The flow velocity of the basilar artery and the left intracranial VA could be reduced by rightward head rotation (Table 1). T1, T2 and fluid-attenuated inversion recovery magnetic resonance imaging (1.5 Tesla system, Picker Edge Eclipse, Picker 98 International, USA) did not reveal any abnormalities. TOF MRA (TR/TE/excitations: 29/6/1) revealed a defect in the posterior circle of Willis (Fig 2A). The right VA was absent, and the left VA (diameter 2.1 mm) supplied the basilar artery (diameter 3.1 mm), which was deviated rightward to 9.8 mm (Fig 2B). Diffusion-



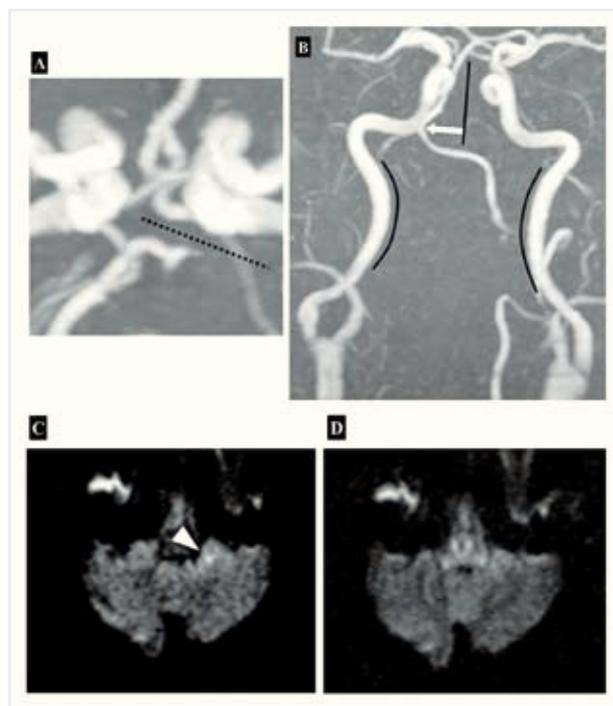
**Fig 1.** A: Leftward nystagmus is noted over right Dix-Hallpike test. B: Pursuit electronystagmogram shows mild, unsmooth eyeball movement. Many spike waves are noted in the eyeball velocity phase. C: Rightward optokinetic nystagmus electronystagmogram (Jung type) is more prominent than leftward. D: An electronystagmogram caloric test shows visual suppression cannot effectively suppress the slow-velocity phase of both caloric nystagmus.

weighted imaging (TR/TE/excitation: 8374/148/1) showed that the left flocculus was hyper-intense (Fig 2C). All blood examinations were within normal ranges.

Therefore, an antiplatelet, 100 mg of aspirin, was given daily and he was asked not to rotate his head rightward up to 90°. In the following month, the symptom occurred one or two times per week, so the medication was halted. Diffusion-weighted imaging (TR/TE/excitation: 8374/148/1) showed the hyper-intensity of the left flocculus had decreased (Fig 2D). The symptom occurred one or two times per month over the following year.

## DISCUSSION

When he was symptomatic, the leftward horizontal nystagmus meant a predominance of the left vestibular function (Fig 1A). There was no other neurological focal deficit. Because vertebrobasilar artery insufficiency or transient ischemic attack are defined as having at least two neurological focal deficits, neither of them were likely to be the cause. Despite a positive Dix-Hallpike test, the characteristics of nystagmus on the Dix-Hallpike test did not indicate a classical benign paroxysmal positional vertigo. In addition, the Epley's canalith repositioning procedures failed to cure his symptom and, therefore, indicated that benign paroxysmal positional vertigo was not the cause. Tests for hyperventilation and a squat to standing position were positive, implying cerebral hemodynamic change was the likely cause, but dysautonomia was unlikely because



**Fig 2.** A: Time-of-flight magnetic resonance angiogram shows a defect in the left side of the posterior circle of Willis (dotted line). B: The basilar artery deviates leftward (hollow arrow). Right vertebral artery is absent. Both extracranial segments of the internal carotid arteries have tortuosities (black curves). C: The diffusion weighted-imaging shows a spot of high intensity in the left cerebellar flocculus (hollow arrow-head). D: The follow-up diffusion weighted-imaging shows the intensity of left cerebellar flocculus has decreased.

**TABLE 1.** Color-coded duplex sonography (EnVisor, Philips, USA) of his vertebrobasilar artery.

	Leftward head rotation				Rightward head rotation			
	EC	IC		BA	EC	IC		BA
	Rt VA	Rt VA	Lt VA		Lt VA	Rt VA	Lt VA	
Diameter (mm)	1.81	*	*	*	2.94	*	*	*
Mean velocity (cm/sec)	*	*	35.2	34.1	38.1	*	16.2	15.2
Mean flow (ml/min)	*	*	*	*	155.2	*	*	*
Resistance index	*	*	0.73	0.58	0.71	*	0.66	0.59
Pulsatility index	*	*	1.14	0.81	1.13	*	0.99	0.87

The reference range of resistance index is less than 0.75, and that of pulsatility index is less than 1.20. EC = extracranial; IC = intracranial; Rt = right; Lt = left; VA = vertebral artery; BA = basilar artery; \* = not measurable.

the orthostatic hypotension test was negative. Electronystagmogram for pursuit (Fig 1B), optokinetic nystagmus (Fig 1C) and the caloric visual suppression test (Fig 1D) showed abnormality, which implicated an impairment of the flocculus.

A color-coded duplex sonogram was manipulated over one side of the temporal and occipital planes when the examinee's head was rotated to the contralateral side, so explicating a stable hemodynamic condition of the VAs and the basilar artery. There was no normal data for the velocity or flow, but the reference range of resistance index was less than 0.75, and that of pulsatility index less than 1.20. Also, the sensitivity and specificity of the duplex sonography for VAH were 20.5% and 90.9% respectively,<sup>3</sup> so it became necessary to analyse the duplex sonogram data in relationship to other image studies, such as an angiogram. It was not possible to evaluate the right extracranial and intracranial VA with a duplex sonogram, and the right VAH was impressed and so an angiogram was scheduled to verify it.

Although a digital subtraction angiogram is the gold standard for diagnosing a vessel disease, it has a small morbidity associated with mortality, so it is impractical and expensive to apply this study to those who have simple dizziness and have no other neurologic deficits. In comparison to a digital subtraction angiogram, the sensitivity and specificity of TOF MRA for a steno-occlusive disease is 94.7% and 97.3% respectively.<sup>4</sup> TOF MRA is indeed an effective alternative to the digital subtraction angiogram, so as far as convenience, economics and safety are concerned, so TOF MRA with color-coded duplex sonography is preferred under the current rules of the Taiwan health security.

Among 18-89 year old Taiwanese adults, 74.6% have defects of the posterior circle of Willis, and among them, 27.8% have an absence of the unilateral posterior communicating artery,<sup>5</sup> with limited circulatory communication between the anterior and posterior circulation (Fig 2A).<sup>6</sup> However, Chen et al., did not mention if the VAH is a co-incident finding of an absent unilateral posterior communicating artery or not.<sup>5</sup> Because of the right VAH, the left VA mainly supplied the lower basilar artery (Fig 2B) and asymmetric VA blood flow might have twisted the basilar artery.<sup>7</sup> The color-coded duplex sonogram revealed the mean velocity of the basilar artery decreased with rightward head rotation, when compared to leftward head rotation (Table 1).

The right VAH is presumed to result from maldevelopment of the vertebrobasilar artery in the 5<sup>th</sup> embryological week. Hypertension or age related atherosclerosis gradually restricts the compliance of the vertebrobasilar artery. Because cervical radiographs showed

small osteophytes, the atlanto-axis was considered anatomically causative. We suggest, therefore, that the atlanto-axis mechanically obstructed the left VA when the patient rotated his head rightward up to 90°. Hemodynamic decrease of the basilar artery (Table 1) led to a hypoperfusion of the posterior-circulation territory, especially the left flocculus (Fig 2C). The left flocculus was transiently injured and failed to modulate the left vestibular nucleus by inhibition; thus, left vestibular function predominated and leftward nystagmus occurred when he was symptomatic. In conclusion, one-side head rotation related episodic vertigo is attributable to VAH.

We could do nothing for the patient's central vascular anomalies. He was asked to change his life-style, and not to rotate his head rightward up to 90°. An antiplatelet was also recommended, diminishing the risk of ischemia or thrombus over the following month. The symptom recurred less frequently than before, and did not bother him any more. In Taiwan, it is seen as controversial to repeat a magnetic resonance image/angiogram for vertigo sufferers, unless a central lesion is confirmed. In this case, the magnetic resonance image/angiogram showed a possible lesion in the left flocculus, so after the symptom subsided, we repeated the image study not only for a medical recheck, but also for academic research. The follow-up diffusion-weighted imaging showed the hyper-intensity of the left flocculus had decreased (Fig 2D). Afterwards, simple life-style changes were recommended. In conclusion, episodic vertigo might be related to unilateral VAH.

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