Case Report

Cervical Posterior Spinal Artery Syndrome Caused By Spontaneous Vertebral Artery Dissection: Two Case Reports and Literature Review

Futao Chen, MMed,^{*,1} Xuemeng Liu, MMed,^{†,1} Tiantian Qiu, PhD,[‡] Chunxue Jia, MMed,^{*} Min Liu, MMed,[‡] Qianxiu Jin, MMed,[‡] Peihong Gao, PhD,[§] and Xiaodong Li, PhD[‡]

Herein, we described 2 patients with posterior spinal artery syndrome (PSAS) caused by vertebral artery dissection. The patients complained of sudden neck pain or walking instability. Neurological examination revealed sensory loss, muscle weakness, and sensory ataxia. Angiography showed double lumen sign or intimal flap in the vertebral artery. T2-weighted imaging and diffusion-weighted imaging of MRI showed a hyperintense lesion in the dorsal side of the cervical spinal cord at different times after onset. Both patients had good outcome after antiplatelet therapy and physiotherapy. A review of previously reported PSAS cases was also conducted in order to improve the understanding and awareness of this rare myelopathy.

Key Words: Vertebral artery dissection—posterior spinal artery syndrome magnetic resonance image

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Introduction

Compared to brain infarction, spinal cord infarction is rare, accounting for 1.2% of all strokes.¹ Spinal cord infarction is usually localized in the anterior spinal artery area, which is known as anterior spinal artery syndrome. Infarction of the posterior spinal artery (PSA) is called PSA syndrome (PSAS), and is very rare. Williamson.² first

2226785371@qq.com. ¹Contributed equally. 1052-3057/\$ - see front matter © 2019 Elsevier Inc. All rights reserved. https://doi.org/10.1016/j.jstrokecerebrovasdis.2019.104601 reported an autopsy case of PSAS in 1895. Due to the lack of characteristic symptoms and radiological features, accurate diagnosis of the disease is difficult for the clinicians and radiologists. We reported 2 PSAS cases caused by vertebral artery dissection (VAD), and reviewed previously published clinical and radiological reports of PSAS.

Case One

A 44-year-old woman was admitted to the hospital with complaints of sudden dizziness and nausea. The patient had no risk factors of cerebrovascular diseases such as smoking, arterial hypertension, hyperlipidemia, and diabetes. On admission, her blood pressure was 130/ 80 mm Hg and heart rate was regular. Neurological examination showed that the patient could speak fluently, and had clear consciousness. However, her bilateral deep tendon reflexes were weak. More serious findings were that deep feeling and tactile sensation on both sides were weak, and Romberg sign was positive. Laboratory examination showed no obvious abnormalities. After 13 hours of the onset of the symptoms, T2-weighted imaging (T2WI) and diffusion-weighted imaging (DWI) showed a

From the *Graduate School, Shandong First Medical University & Shandong Academy of Medical Science, TaiAn, China; †Department of Neurosurgery, FeiXian People Hospital, FeiXian, China; ‡Department of Radiology, LinYi People's Hospital, LinYi, China; and §Department of Radiology, JiNing First People's Hospital, JiNing, China.

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Address correspondence to Xiaodong Li, Peihong Gao, Department of Radiology, LinYi People's Hospital, LinYi, China. E-mail:

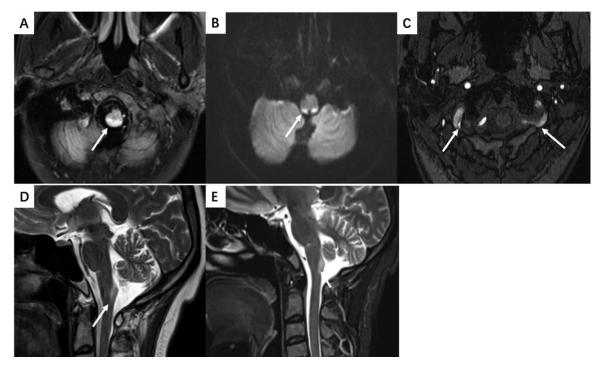


Figure 1. Serial images of bilateral PSAS caused by VAD. (A and B) T2WI and DWI of brain showed a hyperintense lesion in the dorsal side of the medulla; (C) MRA indicated that the bilateral vertebral arteries were stenosis and had the double chamber; (D and E) Sagittal T2WI showed the hyperintense lesion involved spinal cord in the posterior regions at C1 level. Abbreviations: DWI, diffusion-weighted imaging; MRA, magnetic resonance angiography; PSAS, posterior spinal artery syndrome; T2WI, T2-weighted imaging; VAD, vertebral artery dissection.

hyperintense lesion in the dorsal side of the medulla (Fig 1, A,B). Sagittal T2WI showed that the hyperintense lesion involved the spinal cord in the posterior regions at C1 level (Fig 1, D). Cervical MRI was subsequently performed, which showed the same hyperintense lesion at the spinal dorsal side with medulla (Fig 1, E). Magnetic resonance angiography demonstrated stenosis and double lumen sign in the bilateral vertebral arteries (Fig 1, C). Therefore, the patient was diagnosed with bilateral VAD. Combined with laboratory examination, clinical symptoms, and imaging characteristics, the diagnosis of bilateral PSAS caused by bilateral VAD was valid. The patient's symptoms improved significantly after antiplatelet aggregation, with improvement in brain circulation after treatments with mecobalamine and vitamin B6 for 10 days. After 1-month follow-up, the patient's clinical symptoms gradually disappeared.

Case Two

A 34-year-old man complained of sudden numbness of the left limb without obvious reasons for 1 day. He had no other diseases except a history of hyperlipidemia. Laboratory examination showed a major increase in total and low-density lipoprotein cholesterol (300 and 200 mg/dl, respectively). The neurological findings indicated paralysis of left limb, and disturbances of touch and temperature sensation of left lower limb. Computer tomography examination revealed no hematoma or infarction in the brain. However, brain magnetic resonance angiography indicated limitative stenosis of the left vertebral artery. T2WI and DWI showed a hyperintense lesion in left dorsal side of the medulla (Fig 2, A,B,C). Cervical vertebral MRI showed a hyperintense lesion in the posterior regions from the C1-C3 cord on T2WI and fluid-attenuated inversion recovery images (Fig 2, D). Gadolinium-enhanced T1-weighted imaging (T1WI) revealed that this lesion was only slightly enhanced (Fig 2, E). Moreover, double lumen sign and intimal flap of the left vertebral artery were seen on axial T1WI (Fig 2, F). The findings confirmed the diagnosis of left VAD. Therefore, we concluded that the left PSAS was caused by left VAD. The patient received antilipidemic therapy and antiplatelet therapy with rosuvastatin and aspirin. After 1-month follow-up, his symptoms gradually improved. Two months after discharge, he could walk independently.

IRB statement: The patients described in these case reports gave their informed consent. Institutional review board approval of our hospital was obtained for this study.

Discussion

Herein, we reported the clinical and radiological manifestations in 2 patients with PSAS caused by VAD. Additionally, relevant reports of PSAS with complete radiological and clinical details were identified from 1990 to 2019. A total of 22 articles reporting 28 cases of PSAS were identified, with 7 reports of PSAS caused by VAD,



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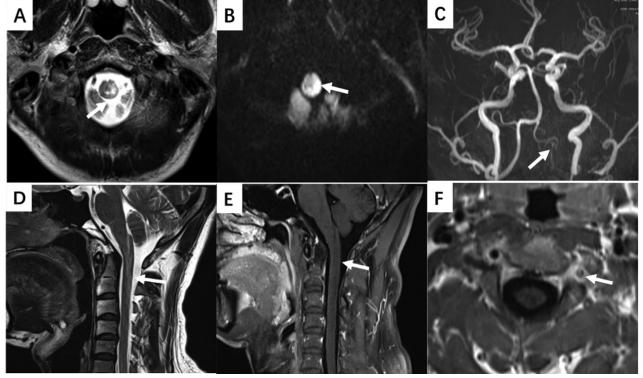


Figure 2. Serial images of left PSAS caused by VAD. (A and B) T2WI and DWI showed a hyperintense lesion in left dorsal side of the medulla; (C) MRA of his brain revealed limitative stenosis of the left vertebral artery; (D) T2WI of his cervical vertebra showed a hyperintense lesion in the posterior regions from the C1-C3 level; (E) Gadolinium enhancement on T1WI indicated this lesion was only slightly enhanced; (F) Axial T1WI showed "double lumen" and "intimal flap" of the left vertebral artery. Abbreviations: DWI, diffusion-weighted imaging; MRA, magnetic resonance angiography; PSAS, posterior spinal artery syndrome; T1WI. T1-weighted imaging; T2WI, T2-weighted imaging; VAD, vertebral artery dissection.

including the 2 cases presented herein.³⁻²⁴ Clinical and radiological details of the 28 cases of PSAS are summarized in Tables 1-3. There are several reasons why spinal cord infarction is rare as compared to brain infarction: (1) the blood vessels supplying the spinal cord are not susceptible to atherosclerosis; (2) spinal tissue oxygen requirement is lower than brain tissue; (3) spinal cord has a rich network of anastomotic vessels.²⁵ These features are more obvious in the PSA than the anterior spinal artery. Moreover, the posterior column receives the lowest blood flow among all tracts in the spinal cord. Therefore, PSAS is less frequently involved than anterior spinal artery syndrome.²⁶

The 2 most common etiologies of PSAS are atherosclerosis and VAD. In the 2 cases reported herein, angiography indicated double lumen sign or intimal flap in the vertebral artery. Double lumen sign and intimal flap are the basis of confirming the diagnosis of VAD.²⁷ Hence, we concluded that PSAS in the 2 patients was caused by VAD. The posterior one-third region of the spinal cord is supplied by 2 PSAs, which are fed by the vertebral artery in the cervical spinal cord.¹⁶ This is the main reason that the lesion of PSAS caused by VAD is localized in the posterior one-third region of the spinal cord. VAD will reduce blood flow of the PSA, which may be the main pathogenesis of PSAS caused by VAD in the cervical spinal cord. VAD could occur due to trauma or spontaneity. A study found that approximately 40% of patients with cervical VAD had a history of cervical trauma in the previous month.¹⁸ However, the 2 cases reported herein had no history of cervical trauma. Therefore, their VAD may be spontaneous.

PSAS mainly involves infarction in the posterior onethird region of the spinal cord, which consists of posterior funiculus and partial lateral funiculus of white matter, and posterior horn of gray matter.²⁴ Fasciculus gracilis, fasciculus cuneatus, posterior spinocerebellar tract, and lateral corticospinal tract are localized in these regions. Therefore, pyramidal tract sign, cerebellar ataxia, or superficial and deep hypesthesia are the main clinical features of cervical PSAS. However, the size and length of lesions are different in the reported cases, so PSAS has no distinct boundaries.¹⁸ The clinical features of PSAS are nonspecific, which hinders the accurate diagnosis of this disease.

Modern imaging techniques not only facilitate accurate diagnosis but also evaluation of any improvement or deterioration to assist the clinicians in making therapeutic interventions.²² The radiological features of patients with PSAS and PSAS caused by VAD are shown in Tables 1 and 3, respectively. Among all the reported patients with PSAS, 16 (57%) patients had lesions in the cervical spine

 Table 1. Previous clinical and radiological Literature of PSAS

Clinical and diagnostic features	No. (%)		
Total cases	28		
Age > 50	17 (61)		
Median age, years (range)	56 (19-84)		
Man	17 (61)		
Risk factors			
Smoking	8 (29)		
Arterial hypertension	14 (50)		
Hyperlipidemia	9 (32)		
Diabetes	3 (11)		
Others	4 (14)		
Etiology			
Atherosclerosis	12 (43)		
Artery dissection	7 (25)		
Iatrogenic	3 (11)		
Trauma	2(7)		
Patent foramen ovale	1(4)		
Other	3 (11)		
Neurological deficits			
Sensory loss	28 (100)		
Muscle weakness	22 (79)		
Sensory ataxia	7 (25)		
Bowel/bladder dysfunction	14 (50)		
MRI findings			
Infarct lesion			
Bilateral	14 (50)		
Unilateral	14 (50)		
Right	7/14 (50)		
Cervical cord location	16 (57)		
Thoracic cord location	12 (43)		
Hyperintensity on T2WI	28 (100)		
Gadolinium enhancement	7/15 (47)		

Abbreviations: PSAS, posterior spinal artery syndrome; T2WI, T2-weighted imaging.

and 12 (43%) had lesions in the thoracic cord. The thoracic-lumbar region was the most common location for PSAS.²⁴ The reason for differences with our results may be that periprocedural spinal cord infarction that is predominantly thoracic was excluded.²⁸ The infarct lesions are mainly located in the upper cervical spinal cord in patients with PSAS caused by VAD due to 2 main reasons. First, in the cervical region of the spinal cord, the upper 3 to 5 cervical spinal arteries are derived from the segmental branches of the vertebral arteries, whereas the lower cervical spinal arteries receive compensatory blood from segmental branches of the ascending and deep cervical arteries and the supreme intercostal artery.²⁹ Second, VAD in most of the included cases occurred in the upper cervical region. T2WI in MRI of all patients (100%) showed a hyperintense lesion in the dorsal side of the spinal cord at different times after onset. The examination of gadolinium enhancement on T1WI had limited value in the diagnosis of PSAS. DWI is the best MRI sequence to reveal early spinal cord infarction.³⁰ High signal with low apparent diffusion coefficient values, which occurs within a few hours after the onset, is a potentially useful and feasible demonstration for PSAS.⁶ In our patients, DWI showed a hyperintense lesion after 13 hours of clinical onset. To the best of our knowledge, this is the first report in which DWI showed a hyperintense lesion in the shortest time between symptom onset and MRI examination. Although PSAS has the above-mentioned radiological manifestations, other spinal cord diseases could have similar imaging features as PSAS. Neuromyelitis optica should be considered as the main differential diagnosis of PSAS. Longitudinal widespread spinal lesions are normally detected on MRI in patients with neuromyelitis optica.¹⁸ Moreover, the initial scans of PSAS may appear

Table 2. Previous clinical features literature of PSAS caused by VAD

Authors	Report years	Age/ sex	Risk factors	First symptom	Neurological deficits	Treatment	Follow-up time	Prognosis
Gutowski et al	1992	35/M	Past smoking	Neck pain	Sensory loss; muscle weakness; bowel/bladder dysfunction	Heparin; physiotherapy	14 w	well
Bergqvist et al	1997	19/M	Smoking	Neck pain	Sensory loss; muscle weakness; Sensory ataxia;			NO
Wang et al	2011	37/M	NO	Acute right neck pain	Sensory loss; muscle weakness; bowel/bladder dysfunction			well
Murata et al	2012	63/F	Smoking hypertension	Neck pain	Sensory loss; muscle weakness; sensory ataxia; bowel/bladder dysfunction			well
Richard et al	2013	30/M	Hypertension	Persistent headache	Sensory loss; muscle weakness; sensory ataxia	· • •		Well
Present cases	2017	44/F	NO	Walking instability	Sensory loss; sensory ataxia	Aspirin; physiotherapy	1 mo	Well
	2018	34/M	Hyperlipidemia	Neck pain	Sensory loss; muscle weakness;	Aspirin; rosuvastatin	2 mo	Well

Abbreviations: PSAS, posterior spinal artery syndrome; VAD, vertebral artery dissection.

NO, not mentioned or not implemented.

Authors	Report years	Location of VAD	Location of lesion	Interval time	Signal change of MRI	Enhancement	Diagnosis styles of VAD
Gutowski et al	1992	Left, C1-C3	Left, C1-C3	8 d	Hyperintensity on T2WI	Peripheral	NO
Bergqvist et al	1997	Right, C1-C2	Bilateral, C2-C3	12 d	Hyperintensity on T2WI	Marked	MRA
Wang et al	2011	Left, C1-C2	Left, C1-C2	2 d	Hyperintensity on T2WI and DWI	NO	MRA
Murata et al	2012	Right, C4-C5	Bilateral, C4-C7	14 d	Hyperintensity on T2WI	Partial	MRA
Richard et al	2013	Right, C1-C2	Right, C2-C3	NO	Hyperintensity on T2WI and DWI	NO	CTA
Present cases	2017	Bilateral, C1	Bilateral, C1	13 h	Hyperintensity on T2WI and DWI	NO	MRA
	2018	Left, C1-C2	Left, C1-C3	1 d	Hyperintensity on T2WI and DWI	Slight	MRA

 Table 3. Previous radiological literature of PSAS caused by VAD

Abbreviations: MRA, magnetic resonance angiography; PSAS, posterior spinal artery syndrome; VAD, vertebral artery dissection.

Interval time, time intervals between symptom onset and the MR examination; NO, not mentioned or not implemented.

normal. Therefore, extensive and serial MRI is a potential necessity if PSAS is suspected.²¹

In summary, the patients with younger age, acute disease course within hours, pain at onset, motor and sensory deficits, and lesions in dorsal side of the upper cervical cord should be highly suspected of PSAS caused by VAD. Early diagnosis of the disease is important for guiding patient management and rehabilitation. Modern imaging techniques are important tools for diagnosing PSAS. However, accurate diagnosis of PSAS remains a challenge for clinicians and radiologists. Therefore, studies with more cases of PSAS describing the clinical and radiological features need to be conducted to improve the understanding and awareness of this rare myelopathy.

Conflict of Interest

All study costs were covered by LinYi People's Hospital.

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