

Bilateral Vertebral Artery Dissection Caused by Atlantoaxial Dislocation

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Abstract

Craniovertebral anomalies are rare causes of vertebral artery dissection. Therefore, appropriate evaluation is necessary in vertebrobasilar strokes, specially in the young. This is particularly true for patients with vertebral stroke without risk factors. Results: Here we report a 24-year-old male presenting with posterior circulation infarcts. The patient had no obvious vascular risk factors. However, digital subtraction angiography showed chronic dissection of both vertebral arteries with partial recanalization. Further evaluation with computed tomogram of neck revealed atlantoaxial dislocation. This is a rare case in which atlantoaxial dislocation has resulted in bilateral vertebral artery dissection and stroke in young.

Introduction

Vertebrobasilar ischemic stroke comprises one-fifth of all ischemic strokes.¹ Dissection of vertebral arteries is an important treatable cause. Though most series attribute arterial dissection to trauma,²⁻⁴ craniovertebral junction (CVJ) anomalies can rarely cause vertebral artery dissection (VAD).^{5,6} Here we report a rare case of bilateral VAD caused by atlantoaxial dislocation (AAD) resulting in posterior circulation stroke.

Case Report

A 24-year-old student presented with intermittent fever, vomiting and malaise for 2 months. A week later, he developed vertigo, diplopia, recurrent vomiting and gait unsteadiness and lapsed into altered sensorium. He gradually recovered with residual dysarthria and bilateral limb incoordination. Historically, no seizures, headache, neck pain, preceding trauma or transient ischemic attacks were noted. He was vegetarian, non-smoker and non-alcoholic. He did not have hypertension, diabetes, hyperlipidemia or cardiac illness. Family history was non-contributory.

Examination revealed normal peripheral pulses and blood pressure. On cardiovascular examination, apical pulse was hyperdynamic with end-diastolic murmur in aortic area. He had ill-sustained attention and hypovolemic speech with decreased output. Fundus was normal. Left side was hypotonic, and right was mildly spastic. Power in all limbs was normal. There were bilateral cerebellar signs. Deep tendon jerks were brisk on the left side and both plantar responses were upgoing. The patient could not walk unsupported.

Routine hematological tests and serum homocysteine were normal. There was hypercholesterolemia and hypertriglyceridemia. HIV serology and antiphospholipid antibodies was negative. Cerebrospinal fluid examination was normal and CSF VDRL was negative. Electrocardiogram was normal while echocardiogram showed mild aortic regurgitation with normal left ventricular function. Doppler of neck vessels revealed total occlusion of right vertebral artery (VA). Computed tomogram (CT) of head showed multiple infarcts in bilateral posterior inferior cerebellar and posterior cerebral arteries territories. MR imaging of brain showed multiple infarcts in the same distribution (Fig. 1 a, b). In addition, CVJ anomaly in the

form of atlantoaxial dislocation with spinal cord compression was noted (Figure 1 c). CT scan of CVJ also revealed AAD with better delineation of the rotation and displacement of the odontoid process (Figure 1 d). Intracranial portions of both VA were poorly visualized on MR angiogram. Thin linear flow was noted in the VAs with irregularity in the vessel wall at C1-2 level (better seen in the left VA) with good flow proximal and distal to it, denoting a localized dissection (Figure 2 a, b). However, no direct evidence of dissection such as intramural blood, intimal flap or double lumen was noted. Hence, digital subtraction angiography (DSA) was done, which was suggestive of chronic dissection of both VA with partial recanalization (Figure 2 c,d). The patient recovered with anticoagulants and gait rehabilitation followed by C1-C2 fusion with sublaminar wires and rib-bone graft.

Discussion

Stroke in vertebrobasilar territory accounts for 20% of all ischemic strokes.¹ Dissection of VA is one of the important causes of vertebrobasilar insufficiency. CVJ anomalies is rarely cause of VAD, but need to be evaluated as they are potentially treatable. A very rare presentation of bilateral VAD secondary to severe AAD causing bilateral vertebrobasilar infarctions has been reported here.

VAD can result from significant neck/head trauma, sometimes by trivial trauma or even spontaneously. In a large series, 83% of dissections were temporally related to trauma.² Trivial trauma may include chiropractic neck manipulation, nose blowing, turning head while reversing a vehicle, labor or delivery. Bilateral internal carotid and VA dissections have been documented following chiropractic manipulation.³ Though dissection of intracranial portion of VA due to trauma is rare, acute dissecting infarction after long periods of repeated trivial pressure such as "Shiatsu" has been reported. Connective tissue disorders such as fibromuscular dysplasia, Marfan's syndrome, Ehler-Danlos syndrome and rarely Turner's syndrome can cause spontaneous dissection. In another series, out of 53% dissections with precipitating factors, sporting activity and chiropractic manipulations were the most common (15% and 11% respectively).⁴ However, bilateral VAD presented only in 24%.

CVJ anomalies rarely cause VAD. The present case is unique in its presentation of VAD occurring bilaterally and secondary to AAD. There are two earlier reports associating basilar invagination with VAD.^{5,6} In both reports, basilar impression resulted in VAD and stroke following manipulation; in one following muscle relaxation and cervical hyperextension during intubation⁵ and in another after undergoing treatment for myelopathy with cervical traction.⁶ This is the only CVJ anomaly

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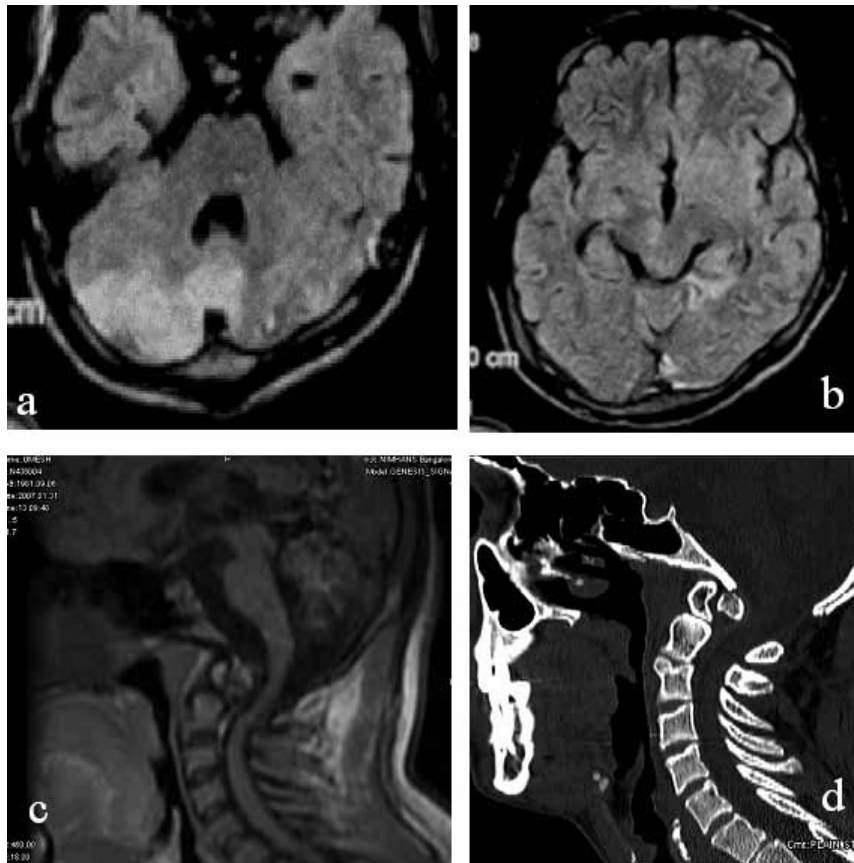


Fig. 1 : MRI FLAIR axial images of brain (a,b) show multiple lesions involving cerebellum, bilateral thalamus, midbrain and left parahippocampal gyrus, suggestive of infarcts; T1-weighted sagittal image of MRI of CVJ (c) shows AAD with cord compression; and sagittal reconstructed CT image of CVJ (d) shows AAD with posterior dislocation of the odontoid process and cervical canal compromise.

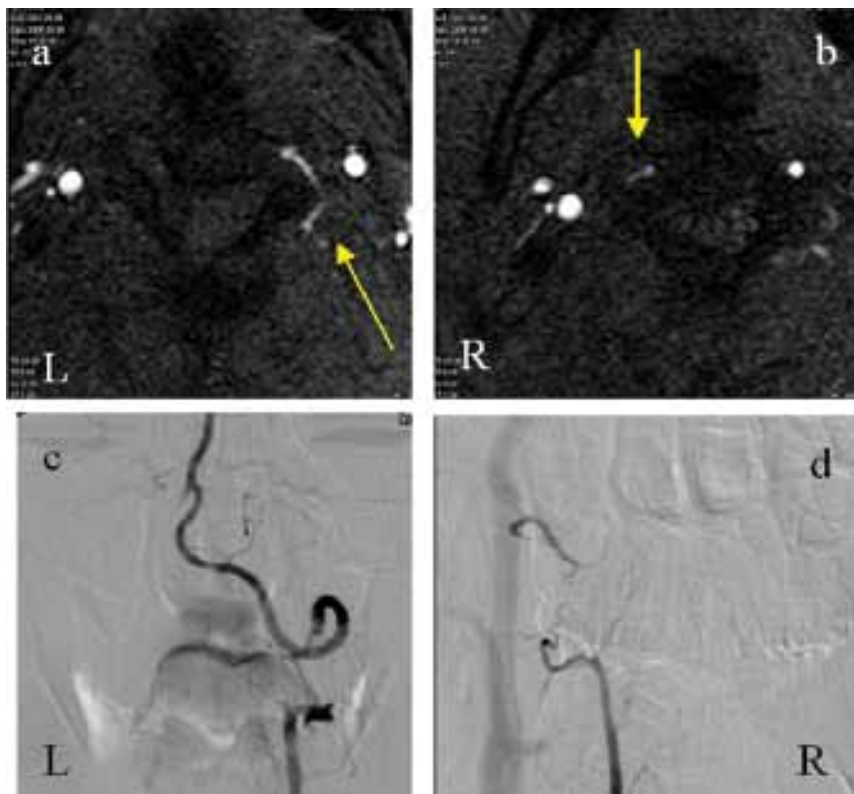


Fig. 2 : MR angiogram images of left (a) and right (b) vertebral arteries shows thin, linear flow with vessel wall irregularities (arrows); and DSA images of left (c) and right (d) vertebral artery injections shows total occlusion of vertebral arteries bilaterally at the level of C1-3 and reformation more distally by muscular anastomotic branches.

which has been reported to cause VAD till date. An extensive search of literature did not show any previous report of VAD due to AAD. Bilateral VAD is hence an extremely rare presentation.

Though 4% of all strokes are attributed to arterial dissection, it is higher (45%) in stroke patients less than 45 years.⁴ Clinical features favoring dissection are neck pain or headache (20-80%), TIAs / ischemic stroke (50-75%) and tinnitus or audible bruit. Of 24 patients with VAD, 16% were bilateral.² The major initial manifestation was occipital or neck pain followed by vertigo and nausea. Headache and/or neck pain was warning sign in 53%, preceding onset of stroke by upto 14 days.⁴ The case discussed here demonstrates the importance of evaluation of recurrent TIAs or stroke in vertebrobasilar territory. Apart from recurrent vertebrobasilar TIAs, the patient did not have neck pain, headache or trauma.

Many consider DSA as diagnostic gold standard.⁶ Angiographic findings include tapered stenosis or occlusion, string sign, pseudoaneurysm, intimal flap, distal pouch formation and evidence of fibromuscular dysplasia. Different studies give varied sites for VAD. Saeed AB et al found C1-C2 level (16 arteries, 51%) as most common region of VA dissection.⁴ Intracranial VAD was found in eight arteries (25%). However, Bartels E noted that most frequent site of VAD (43%) was distal VI and proximal V2-segment (C6 vertebral level).² Interestingly, here MR angiography and DSA showed bilateral V3 VAD with distal reformation by muscular branches of external carotid arteries.

Majority of patients with VAD have a favorable outcome following treatment.⁴ However, poor prognosis is usually associated with bilateral VAD and intracranial VAD accompanied by subarachnoid hemorrhage. The patient described did well despite having bilateral VAD with a compromising AAD. This could be attributed to the chronic nature of dissection with partial reformation of both VA by muscular branches of external carotid arteries. Surgical treatment had further stabilized the craniovertebral region preventing further TIAs or stroke.

Conclusion

CVJ anomalies are an important treatable cause of VAD and should be evaluated for in vertebrobasilar strokes in young. This case highlights the association of AAD with VAD that can be bilateral due to subluxation of the joint and damage to both VAs. Hence, patients with vertebral stroke without obvious vascular risk factors must undergo requisite investigations for assessment of the CVJ. Such timely assessment enables surgical correction and prophylaxis against excessive neck movement that can precipitate strokes.

References

1. Caplan LR. In: Caplan LR, ed. *Caplan's Stroke- A Clinical Approach*. 3rd edition, Boston: Butterworth-Heinemann, 2000.
2. Bartels E. Dissection of the extracranial vertebral artery: clinical findings and early noninvasive diagnosis in 24 patients. *JNeuroimag* 2006;16:24-33.
3. Nadgir RN, Loevner LA, Ahmed T, Moonis G, Chalela J, Slawek K, Imbesi S. Simultaneous bilateral internal carotid and vertebral artery dissection following chiropractic manipulation: case report and review of the literature. *Neuroradiol* 2003;45:311-4.
4. Saeed AB, Shuaib A, Al-Sulaiti G, Emery D. Vertebral artery dissection: warning symptoms, clinical features and prognosis in 26 patients. *Can J Neurol Sci* 2000;27:292-6.
5. Zotter H, Zenz W, Gallistl S, Zohrer B, Lindbichler F. Stroke following appendectomy under general anaesthesia in a patient with basilar impression. *Acta Anaesthesiol Scand* 2000;44:1271 -2.
6. Dickinson LD, Tuite GF, Colon GP, Papadopoulos SM. Vertebral artery dissection related to basilar impression: case report. *Neurosurg* 1995;36:835-8.