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CASE REPORTS

Traumatic vertebral artery dissection mimicking central pontine myelinolysis: A case report

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Abstract

A 27 year-old Chinese man, involved in a motor vehicle accident, presented with rapidly progressive pseudobulbar palsy and spastic tetraplegia. Magnetic resonance imaging (MRI) of the brain showed central pontine T2 hyperintensity with an unaffected outer rim, consistent with central pontine myelinolysis. There was no hyponatraemia before MRI and he was neither an alcoholic nor malnourished. Cerebral angiogram confirmed the diagnosis of right vertebral artery dissection. Vertebral artery dissection should be considered in a case with imaging suggestive of central pontine myelinolysis.

INTRODUCTION

Central pontine myelinolysis (CPM) or osmotic demyelination syndrome, as described by Adams in 1958, is a demyelinating disorder in the centre of basis pontis.[1] It is characterized by rapidly progressive onset of spastic quadriplegia and pseudobulbar palsy, with progression to locked-in syndrome. Magnetic resonance imaging (MRI) of the brain typically shows symmetrical T2-weighted hyperintense focus in the central part of the pons with sparring of the outermost tegmentum and a peripheral rim of ventral pontine tissue. In some cases, two central symmetric isointense structures within the hyperintense pons, representing the spared descending corticospinal tracts, may be observed.[2] It was originally described in hyponatraemic patients with a history of alcohol abuse[1] but more recently has been documented following liver transplantation[3] and in patients with hypokalaemia.[4,5] CPM had been reported in a normonatraemic alcoholic patient with malnourishment.[6-7] An unusual case of CPM in shock, with spontaneous recovery had also been reported.[8] Other rarer causes include psychogenic polydipsia, burns (infrequent, often with hypernatraemia), after pituitary surgery, post-urological/gynaecological surgery, and prolonged diuretics.[9]

Vertebral artery dissection (VAD) is an important cause of brainstem stroke in patient younger than 45 years.[10,11] The locations of the dissections were classified as extradural (V1, V2 and V3) and intradural (V4). The intradural dissection is the commonest site of dissection in an angiographic study of VAD.[12] Patients with intradural dissection usually present with severe headache and subarachnoid haemorrhage, as well as infarction.[12] The most common site for infarction was cerebellum followed by the occipital lobe.[13] In the patients with brainstem stroke, lateral medullary syndrome was the commonest clinical picture.[14] Locked-in syndrome in VAD had been reported but uncommon[15-16], and it is usually related to basilar artery involvement.

This case demonstrated an uncommon presentation of traumatic VAD, which presented with locked-in syndrome and central pontine involvement in neuroimaging without predisposing factors for central pontine myelinolysis. Association between CPM and vertebral artery dissection has not been previously reported in the medical literature.

CASE REPORT

This is a 27 year old Chinese gentleman, who worked in a recreation club. He met with a motor vehicle accident at midnight, back home from work. He was a driver of a car which skidded and overturned and hit a tree. He managed to get out of his car without help and called his family through handphone. He was admitted on the same day and was found to have a fracture of the left body of the mandible and the right condyle, and also fracture of spinous process of C6 and C7.

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He was otherwise well and able to speak to his family though not able to open his mouth due to fracture. He developed right sided hemiplegia and dysarthria on the second day which progressed to tetraplegia the next day. He was ventilated due to respiratory distress, but consciousness was preserved.

MRI brain showed asymmetrical, heterogenous T2-weighted hyperintense focus in the central pons with an unaffected outer rim suggestive of central pontine myelinolysis (Figure 1A). The lesion was contrast enhanced on T1-weighted MRI (Figure 1B). However, there was no documentation of hyponatraemia throughout the admission and liver function test was normal. He had been working in the recreation club for 3 months prior to the accident and had been on regular 5-6 units of alcohol daily since then. However, there was no

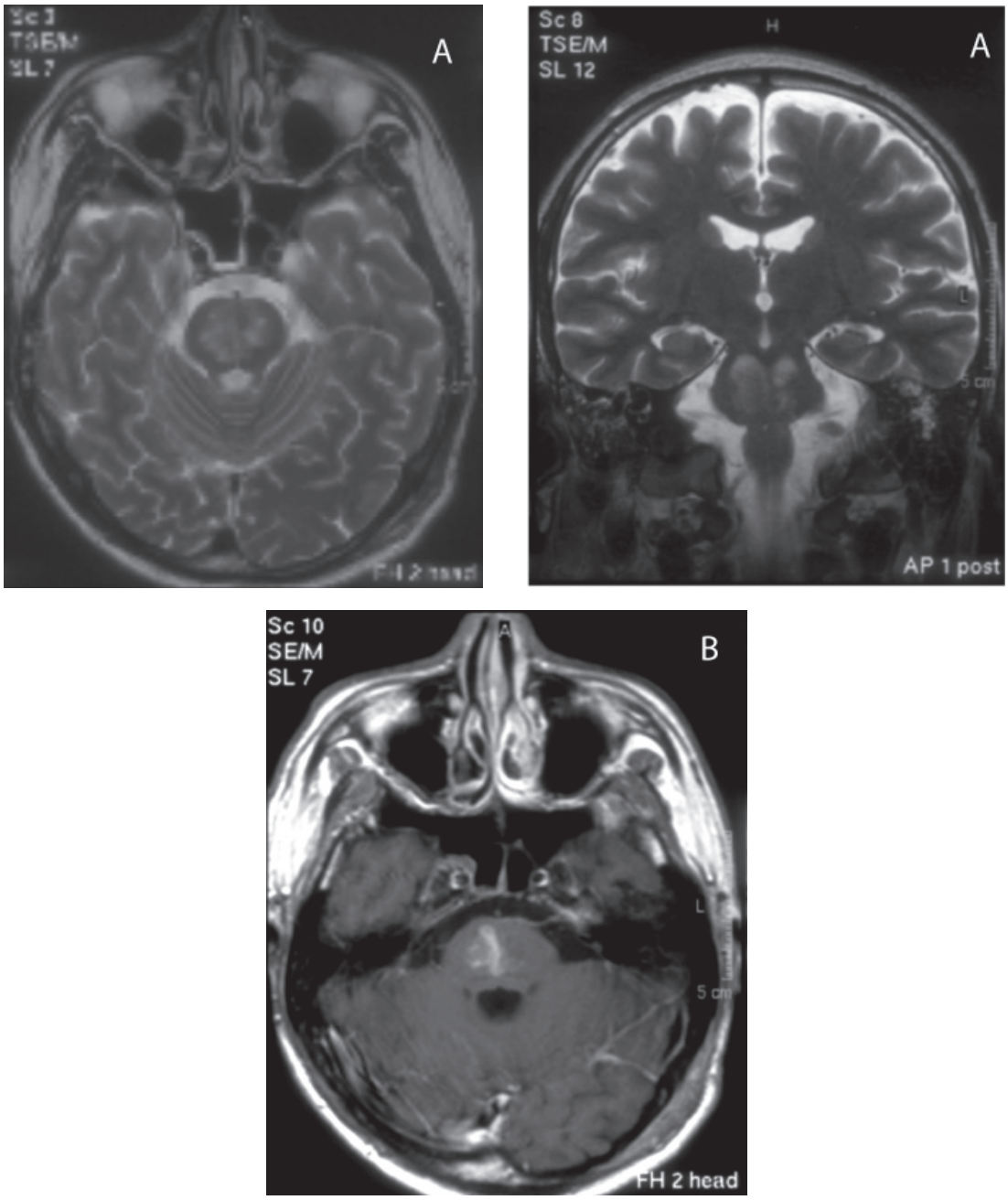

Figure 1 **A**: MRI brain showed asymmetrical, heterogenous T2-weighted hyperintense lesions in the central pons.
B: Post-contrast T1-weighted MRI showing contrast enhancement of the pontine lesion.

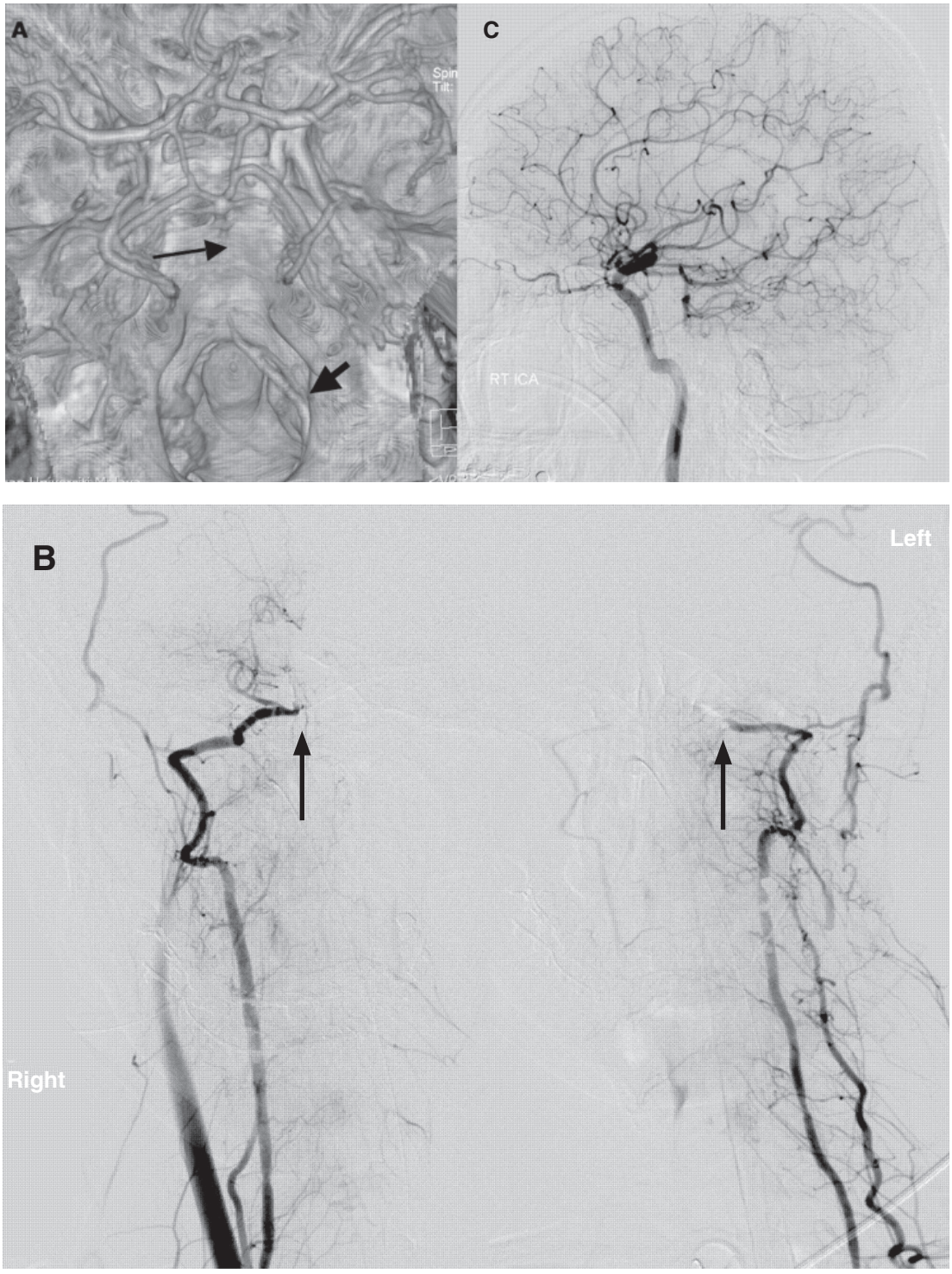

Figure 2 **A**: CTA showed absence of the left vertebral artery at level of foramen magnum. The proximal basilar artery is absent, the distal basilar artery (narrow black arrow) was visualized via backflow from the posterior communicating arteries. Right vertebral artery gives off the posterior inferior cerebellar artery before tapering off (large arrow). **B**: Cerebral angiogram demonstrating the bilateral smooth tapered occlusions of the vertebral arteries at level of skull base (arrows). **C**: Rt ICA run, demonstrating extensive back-flow into the basilar sytem and supplying the cerebellar hemisphere.

clinical evidence of alcohol dependence and no signs of alcohol withdrawal during admission. Clinically, he was a well-built man with no signs of malnourishment.

Upon transfer to our center for further evaluation, he was found to be alert with spontaneous eye opening, able to communicate by eye blinking and head turning. His pupils were equal bilaterally and reactive to light. Nervous system examination showed bilateral facial nerve palsy with pseudobulbar palsy, and spastic tetraplegia.

In view of trauma and absence of predisposing factor for central pontine myelinolysis, a Computed Tomography angiography (CTA) and subsequently cerebral angiogram were performed, demonstrating bilateral vertebral dissections at the foramen magnum (Fig. 2). With rehabilitation, he had partially recovered with ability to swallow and minimal improvement of motor power to grade 1.

DISCUSSION

The clinical features of this case, together with T2-weighted hyperintense focus in the central part of the pons with an unaffected outer rim in the MRI, were suggestive of central pontine myelinolysis. There was no documented hyponatraemia, and the patient was not malnourished. This patient had daily consumption of alcohol for 3-months without overt clinical effect. Most literatures reported CPM in patients with chronic alcohol consumption with alcohol related complications e.g. hepatitis, cirrhosis, pancreatitis and malnourishment.[1, 7, 17] Absence of hyponatraemia and alcohol associated complications makes alcohol consumption as a cause for CPM in this case unlikely.

Pontine infarction, though rare, can be the primary presentation of VAD if there is basilar artery thrombosis or extension of dissection into basilar artery. Infarction in VAD more commonly involves cerebellum, followed by occipital lobe.[13] In the patients with brainstem stroke secondary to VAD, lateral medullary syndrome is the most common clinical picture.[14] However, though rare, there are a few case reports of traumatic basilar artery dissection causing pontine infarction. Pontine involvements in these cases are usually asymmetrical[18] or unilateral[19], associated infarction in the adjacent structures e.g. cerebellum and occipital lobes[20], though involvement of the ventral portion of the pons presenting as lock-in syndrome has also been reported.[21]

Pontine infarction is usually unilateral.[22] Isolated bilateral symmetrical pontine infarction is uncommon, although this can be explained by simultaneous bilateral paramedian artery occlusion. A review of basilar artery occlusion disease showed that out of 87 patients, 5 patients (5.7%) had tetraparesis or tetraplegia, and only one patient with locked-in syndrome.[16] A similar presentation with pathology was reported by Fisher in 1977 but the pontine involvement is multiple, asymmetrical with involvement of the periphery of the pons.[18]

Therefore, the characteristics of centrality in location and sparing of the periphery in the pons, and isolated pontine lesion without involvement of adjacent cerebellum, are points against the diagnosis of pontine infarction. In fact, in Victor and Adams' original article on CPM, he argued on clinical grounds that CPM was unlikely to be an infarct because there was no segmental brain stem signs, and the signs were too limited for a basilar occlusion.[1] In our patient, the angiographic evidence of vertebral artery dissection and basilar artery involvement is strongly supportive of the diagnosis of pontine infarction. Cerebellum and occipital lobes were spared because there were good collateral vessels from middle cerebral artery.

Nevertheless, there are features in our patient's MRI that are atypical for CPM. CPM is classically described as bilaterally symmetrical and predominantly basis pontis involvement as a result of myelin destruction with relative sparing of the neurons and axons.[1] Contrast enhancement was not a consistent feature, and only reported as faint and peripheral in one case report.[23] Thus, though our patient's MRI mimic CPM in its central pontine involvement, is atypical in the asymmetrical and heterogenous distribution of MRI T2-weighted hyperintensity, and prominent contrast enhancement.

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