Neurology Asia 2010; 15(2) : 179 – 183

Pathological laughing with syncope and occipital hypoperfusion as an unusual late effect of pontine infarct

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Abstract

Occipital lobes are not usually implicated in the current proposed pathways of pathological laughter. We present a case of occasional pathological laughter associated with pontine infarct. Brain SPECT revealed hypoperfusion of bilateral occipital lobes in addition to an underlying abnormal regulation of the cortico-pontine-cerebellar pathway. The cause of bilateral occipital hypoperfusion was thought to be due to vertebrobasilar stenosis in our patient. The co-existence of bilateral occipital lobe hypoperfusion in our patient suggest that occipital lobes may also be involved in the generation of pathological laughter.

INTRODUCTION

Pathological laughing and crying is not rare in post stroke patients. The prevalence of pathological laughing and crying in cerebrovascular disease is 11-34 %.[1] But syncope immediately following pathological laughter is rare. We present a patient with an old right pontine infarct with syncope immediately after episodes of pathological laughter. Brain Tc-99m-ethyl cysteinate dimmer (Tc-99m-ECD) single photon emission computed tomography (SPECT) revealed hypoperfusion not only in cerebellum, inferior temporal lobe, but also bilateral occipital lobes. Occipital lobes are currently not in the proposed pathways of pathological laughter.[2]

CASE REPORT

A 65-year-old Taiwanese man visited the Emergency Department of the Mackay Memorial Hospital because of syncope. The syncope developed immediately after abrupt laughter whilst watching his grandson playing a video game. He could not explain why he burst into laughter. The patient subsequently lost consciousness for few seconds, without limb twitching, and recovered within half an hour. There was no strain or sudden change of body posture before syncope.

The patient's history included a right pontine infarct, diagnosed 4 months prior to the onset of the current symptoms.

The neurological examination in the Emergency Department revealed left hemiparesis, a sequel of the previous right pontine infarct. Similar symptoms took place on the next day and two weeks later. But there was no syncope in the third episode of pathological laughter.

Brain MRI (Figure 1) taken 4 months previously revealed a right pontine infarct. Brain MR angiography (Figure 2) showed hypoplasia of the right vertebral artery, segmental narrowing of left vertebral artery, and occluded basilar artery. Transcranial color-coded ultrasonography taken 3 days after MRI found high resistant flow profile in bilateral vertebral arteries suggesting increased downstream vascular resistance such as distal stenosis, occlusion or generalized atherosclerotic change.

The patient was then examined using Tc-99m-ECD SPECT (Figure 3) to evaluate the cause of pathological laughter with syncope. Although neither laughing nor syncope occurred during the scanning, the examination revealed hypoperfusion of the left inferior temporal, bilateral occipital lobes, and bilateral cerebellar hemispheres.

DISCUSSION

The subthalamic nucleus, anterior cingulate, and hypothalamic region are thought to regulate emotional expression.[3-5] The cerebellum modulates the profile of emotional response unconsciously and automatically according to the information it receives from the cerebral cortex.[6] The laughing center locates at the ventral pontomedullary area, which coordinates emotional vocalization, facial expression, and expirations.[7-8] Therefore a

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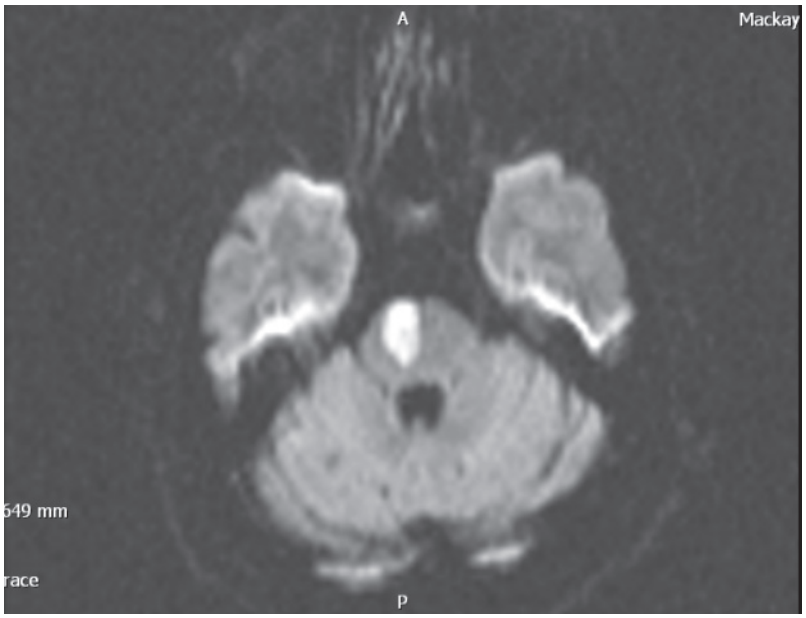

Fig 1 Brain MRI (DWI) revealed a right ventral pontine infarct.

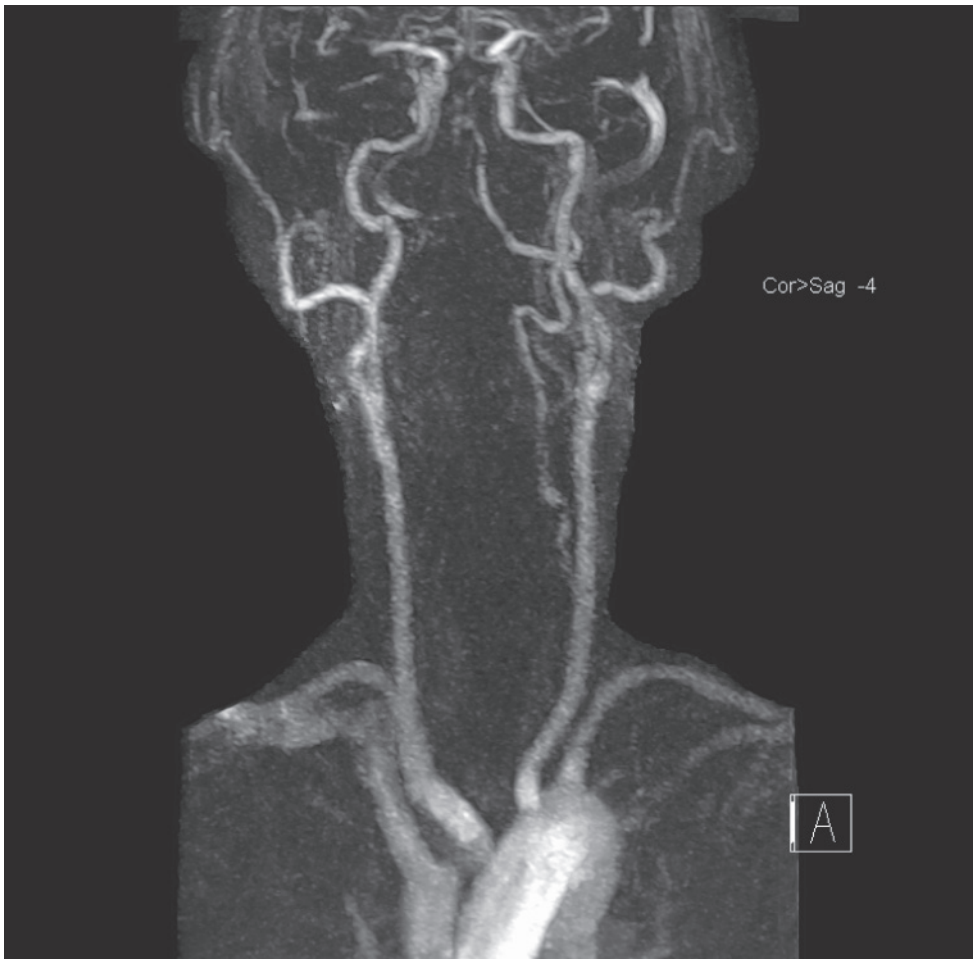

Fig 2. Brain MRA showed hypoplasia of right vertebral artery, segmental narrowing of left vertebral artery, and occluded basilar artery.

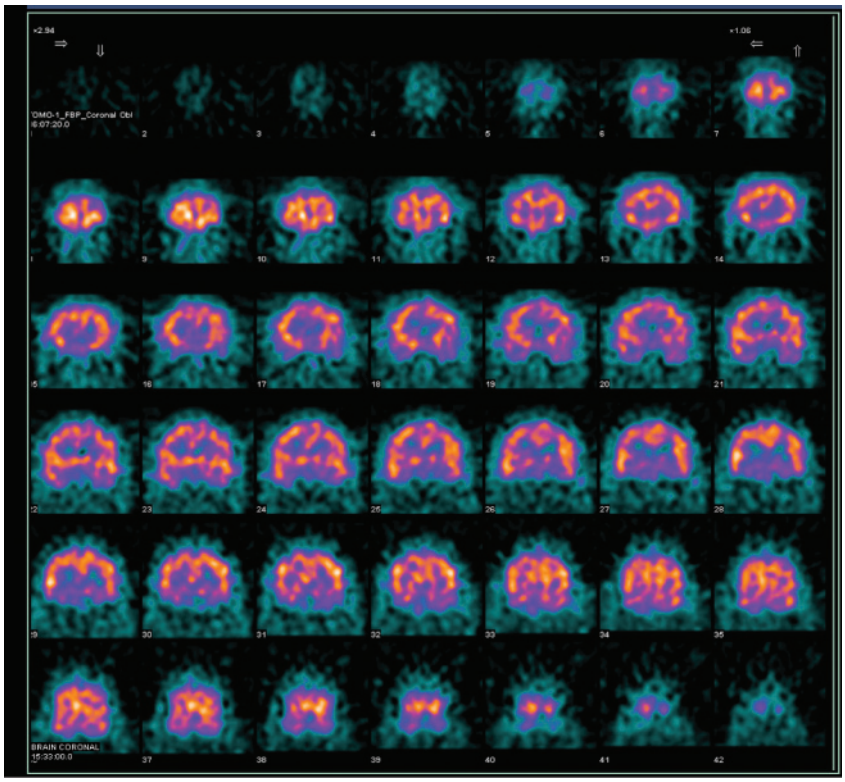

Fig 3a. Coronal view of brain SPECT

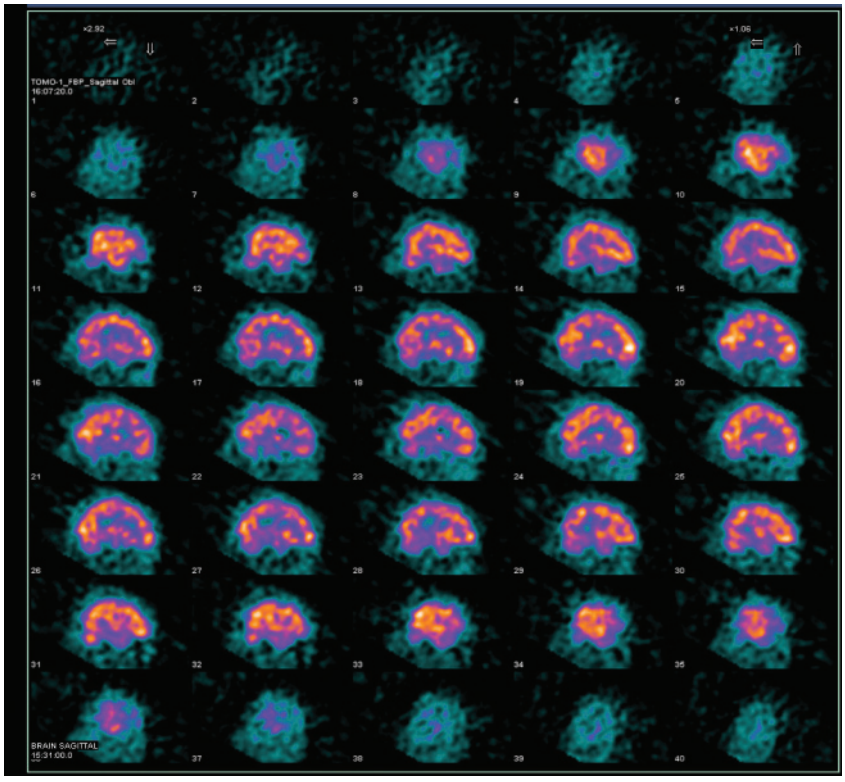

Fig 3b. Sagittal view of brain SPECT

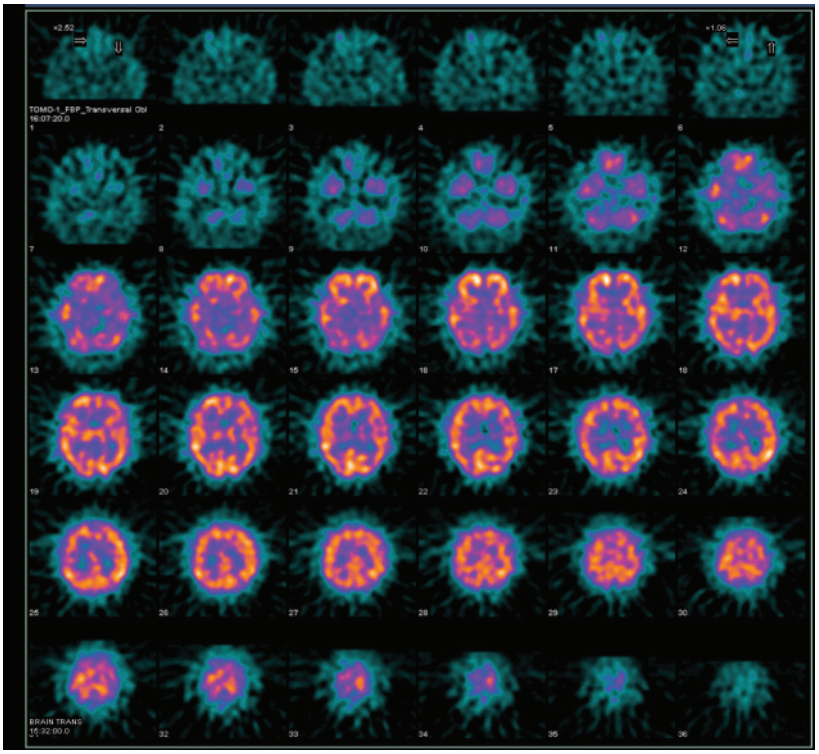

Fig 3c. Transversal view of brain SPECT

Fig 3. The coronal, sagittal, and transversal views of brain SPECT showed hypoperfusion of left inferior temporal, bilateral occipital lobes, and bilateral hemispheres of cerebellum.

proposed pathway associated with pathological laughing and crying involves frontal lobes, brainstem, basis pontis, and cerebellum. The pons, as suggested by its name, bridges cerebrum and cerebellum. In our patient, an infarct of the pons may be implicated in pathological laughter because of interruption of cortico-pontine-cerebellar pathways including the ventral pontomedullary laughing center.[9-10]

Most results of brain SPECT in our patient may be interpreted as interruption of cortico-pontine-cerebellar pathway secondary to pontine infarction[11-13] except bilateral occipital hypoperfusion. The vertebrobasilar stenosis with high resistant blood flow in brain MRA and Doppler studies may explain the bilateral occipital hypoperfusion. The co-existence of bilateral occipital lobe hypoperfusion in our patient suggest that occipital lobe may also be involved in the generation of pathological laughter.

Occluded basilar artery in MRA and high resistance flow profile in transcranial color-coded ultrasonography also implied the development of collateral circulation of vertebrobasilar system. The patient's occasional, not persistent, pathological laughter probably were due to collateral circulation with partial compensation of ischemia.

The laughter-induced syncope is probably due to increased intrathoracic or intra-abdominal pressure via a Valsalva mechanism in association with the laughter.[14]

REFERENCES

1. Parvizi J, Arciniegas DB, Bernardini GL, *et al*. Diagnosis and management of pathological laughter and crying. *Mayo Clin Proc* 2006; 81:1482-6.
2. Parvizi J, Coburn KL, Shillcutt SD, Coffey CE, Lauterbach EC, Mendez MF. Neuroanatomy of pathological laughing and crying: a report of the American Neuropsychiatric Association Committee on Research. *J Neuropsychiatry Clin Neurosci* 2009; 21(1):75-87.
3. Low HL, Sayer FT, Honey CR. Pathological crying caused by high-frequency stimulation in the region of the caudal internal capsule. *Arch Neurol* 2008; 65:264-6.
4. Mallet L, Schupbach M, N'Diaye K, *et al*. Stimulation of subterritories of the subthalamic nucleus reveals its role in the integration of the emotional and motor

aspects of behavior. *Proc Natl Acad Sci U S A* 2007; 104:10661-6.
5. Kahane P, Ryvlin P, Hoffmann D, *et al*. From hypothalamic hamartoma to cortex: what can be learnt from depth recordings and stimulation? *Epileptic Disord* 2003; 5:205-17.
6. Parvizi J, Anderson SW, Martin C, *et al*. Pathological laughter and crying: a link to the cerebellum. *Brain* 2001; 124:1708-19.
7. Wilson SAK: Some problems in neurology, II: pathological laughing and crying. *J Neurol Psychopathol* 1924; 16:299.
8. Poeck K. Pathological laughter and crying. In: Fredericks JAM, ed: Handbook of Clinical Neurology, Vol 45. Amsterdam, Elsevier Science, 1985, 219-25
9. Oh K, Kim HJ, Kim BJ, Park KW, Lee DH. Pathological laughter as an unusual manifestation of acute stroke. *Eur Neurol* 2008;59(1-2):83-4. Epub 2007 Oct 11.
10. Tei H, Sakamoto Y. Pontine infarction due to basilar artery stenosis presenting as pathological laughter. *Neuroradiology* 1997; 39(3):190-1.
11. Samaniego EA, Stuckert E, Fischbein N, Wijman CA. Crossed cerebellar diaschisis in status epilepticus. *Neurocrit Care* 2009; 12(1):88-90
12. Tsuda Y, Ayada Y, Izumi Y, Ichihara S, Hosomi N, Ohkawa M, Matsuo H. Cerebellar diaschisis in pontine infarctions: a report of five cases. *Eur J Nucl Med* 1995; 22(5):413-8.
13. Fazekas F, Payer F, Valetitsch H, Schmidt R, Flooh E. Brain stem infarction and diaschisis. A SPECT cerebral perfusion study. *Stroke* 1993; 24(8):1162-6.
14. Amaki M, Kamide K, Takiuchi S, Niizuma S, Horio T, Kawano Y. A case of neurally mediated syncope induced by laughter successfully treated with combination of propranolol and midodrine. *Int Heart J* 2007; 48(1):123-7.