

Secondary mania in a patient with delayed anoxic encephalopathy after carbon monoxide intoxication

Bon D. Ku ^a, Hee Young Shin ^b, Eun-Joo Kim ^b, Key Chung Park ^c,
Sang Won Seo ^b, Duk L. Na ^{b,*}

^a Department of Neurology, Kwandong University College of Medicine, Myongji Hospital, Gyeonggi, Korea

^b Department of Neurology, Sungkyunkwan University, School of Medicine, Samsung Medical Center, Seoul, Korea

^c Department of Neurology, Kyung Hee University, School of Medicine, Kyung Hee Medical Center, Seoul, Korea

Received 6 July 2005; accepted 19 October 2005

Abstract

Mania is a rare clinical manifestation of delayed anoxic encephalopathy (DAE). Prior case reports on mania after hypoxic injury involved patients with a previous history of mania or depression, potentially reflecting a recurrence of premorbid mood disorders after hypoxia rather than pure secondary mania. Herein, we report a 55-year-old woman with no past history of neurological or psychiatric illness, who developed mania as a symptom of DAE after carbon monoxide intoxication. Brain magnetic resonance imaging showed diffuse white matter lesions, particularly visible in the frontal white matter. This frontal lesion may have prevented frontal inhibition from being transmitted to the basotemporal limbic area, resulting in mania manifested as a burst of limbic activity.

© 2006 Elsevier Ltd. All rights reserved.

Keywords: Delayed anoxic encephalopathy; Secondary mania

1. Introduction

It is known that anoxic brain injury results in acute as well as delayed neurological manifestations. One of the most frequent delayed manifestations is delayed anoxic encephalopathy (DAE), which occurs several days or weeks after full recovery from acute hypoxia.¹ Choi reported that 65 of 2360 patients (2.75%) with acute carbon monoxide (CO) intoxication developed DAE and that the characteristic symptoms in these patients were abulia, akinetic mutism, agitation/confusion, diffuse cortical dysfunction, urinary incontinence, speech disturbance, involuntary movement and depression.¹ Min also reported similar symptoms in 86 patients with DAE after CO intoxication,² but mania was not described as a symptom of DAE in these large series.^{1,2}

Although the main cause of DAE is CO intoxication, DAE also results from other medical conditions, such as shock and hypoxia.^{3,4} To date, there have been three case reports on elevated mood as a symptom of DAE following various medical conditions.^{5–7} Yagnik et al.⁵ reported two patients, who had a history of drug intoxication and depression, who developed DAE after attempting suicide. These patients exhibited an elevated mood that was considered to be a component of the psychotic symptoms they had experienced for a number of years. Collins and

Jacobson reported a patient who had abnormal motor behaviour and elevated mood following anoxia.⁶ However, this patient had bipolar disorder premorbidly, and thus the elevated mood may have been a switch to the manic phase of the premorbid bipolar disorder. Kumar and Agarwal⁷ reported the case of a patient who did not fulfill the mania criteria of the *Diagnostic and Statistical Manual of Mental Illness* (DSM-IV).⁸ Herein, we report on a patient with secondary mania as a symptom of DAE after CO intoxication who had no premorbid history of depression or mania and fulfilled the DSM criteria of mania.

2. Case report

A 55-year-old right-handed woman with a 5-year elementary school education was admitted to the Neurology Department at Samsung Medical Center with memory disturbance and abnormal behaviour for 7 days. Thirty days prior to admission, she had a quarrel with her husband and slept in a closed space containing a charcoal stove, which remained on during this time. The next morning, she was found to have an impaired sensorium and to have urinated on the bed. She soon became alert, but her headache, dizziness and nausea persisted for 1 day. She subsequently continued her normal life until 7 days prior to admission, at which time she became increasingly disoriented to time and place over the next 3 days and exhibited a confused mental state and abnormal behaviour. She was

* Corresponding author. Tel.: +82 2 34103591; fax: +82 2 34100052.
E-mail address: dukna@smc.samsung.co.kr (D.L. Na).

wandering in her village with topographical disorientation. She had difficulty using her cell phone. Other unusual behaviours were soliloquizing incomprehensible words, a misconception that ‘neighbors speak ill of her’, somnolence, intermittent irritability and aggressive behaviour towards her granddaughter and fumbling and touching things around her.

The only remarkable event in her past medical history was a total radical hysterectomy performed 10 years previously, with no history of hypertension, diabetes, cardiac disease, liver disease or neurological or psychiatric disorders.

On admission, the patient was disoriented as to time and place and had a short attention span, scoring only 8/30 on the Korean version of the Mini-Mental State Examination (MMSE). The rest of the examination was negative for neurological abnormalities, including parkinsonism, gait disturbance and frontal lobe releasing signs, but symmetric hyperreflexia and bilateral extensor plantar responses were present.

Investigations, including a complete blood cell count, liver function test, blood urea nitrogen/creatinine, urine analysis, thyroid function, syphilis serology and electrolyte profiles, were unremarkable. The electroencephalogram and cerebrospinal fluid examination showed no abnormal findings. The fluid light attenuation inversion recovery (FLAIR) and T₂-weighted magnetic resonance imaging (MRI) showed diffuse high signal intensities in both the periventricular and deep white matters without contrast enhancement (Fig. 1).

For the first 5 days after admission, the patient remained quiet and abulic, with an expressionless face, but her mood began to become elevated thereafter (35 days post-hypoxia). She laughed frequently and became very talkative, in a manner that was totally unlike her premorbid character

of being shy and quiet. Her speech was fast and incoherent. She inadvertently touched things around her and embraced people carelessly. She took other people’s belongings without permission. She also disturbed other patients or caregivers by talking to them and wandering about at night, sleeping for less than 4 h/day. She ate continuously unless she was prevented from doing so.

To quantify our patient’s mania, we used the Young mania scale 5 days after the appearance of the manic symptoms (40 days post-hypoxia). This scale consists of 11 measures, each being scored from 0 to 8 points, and the score for our patient was 22/88, which is indicative of a moderate degree of mania (12–19, mild mania; 20–25, moderate mania; ≥ 26 , severe mania). On the same day, we also performed the MMSE and the Korean version of the Neuropsychiatric Inventory (NPI).⁹ The MMSE score was unchanged (7/30). The frequency and severity of the euphoria subscale of the NPI were rated as 4/4 and 3/3, respectively. Other subscales rating as positive were agitation, disinhibition, aberrant motor behaviour, night-time behaviour and appetite/eating changes, with the total NPI score being 45/144.

These DAE symptoms, along with the mania, remained largely unchanged for the next 10 days, but began to improve spontaneously without medication until discharge (50 days post-hypoxia). On follow-up examination, at 65 days post-hypoxia, the patient’s abnormal behaviours had disappeared, except for mild irritability and fluctuating mood, and she had partially regained the ability to manage household chores, such as cleaning and washing. At 75 days post-hypoxia, her score on the Young mania scale was 8/88 and her MMSE score was 11/30, with a total NPI score of 8/144 (euphoria subscale: frequency 1, severity 1). At 150 days post-hypoxia, the patient had almost returned to the premorbid state, being able to perform household tasks independently (MMSE score 22/30; Young mania scale 3/88; NPI 1/144).

3. Discussion

Although the patient’s CO haemoglobin level was not measured in the acute phase, her recent history of hypoxia, diffuse cognitive deficit with abnormal behaviour after a lucid interval of 3 weeks and diffuse periventricular high-signal MRI abnormalities are all compatible with the DAE diagnostic criteria proposed by Yagnik and Burns.⁵

Our patient’s euphoric symptoms, which started 35 days after hypoxia (5 days after DAE), included increased psychomotor activity, elevated mood, laughing, hyperphagia, talkativeness, irritability and decreased sleep, which are compatible with the mania criterion proposed by DSM-IV. The quantification of mania by the Young mania scale further supported the presence of a moderate degree of mania. Secondary mania is associated with the use of drugs, such as alcohol, caffeine, sympathomimetics, lisinopril, felbamate, famotidine, diltiazem, steroids, amantadine, bupropion, isoniazid, triazolam and procyclidine, as

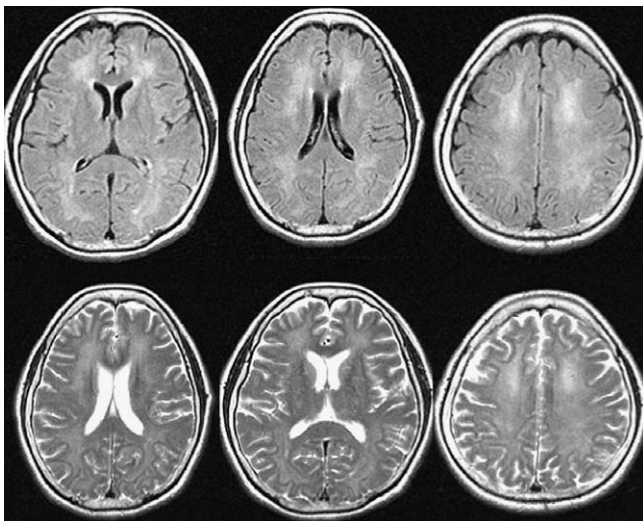


Fig. 1. Brain magnetic resonance imaging (MRI): fluid light attenuation inversion recovery (FLAIR) (upper row) and T₂-weighted (lower row) images showing diffuse confluent high-signal intensity in the subcortical white matter, most prominent in the frontal white matter.

well as various medical conditions, including vitamin B₁₂ deficiency, thyrotoxicosis, HIV infection, traumatic brain injury, brain tumour, cerebral infarction, Creutzfeldt–Jakob disease and multiple sclerosis.^{4,7,10,11} Our patient did not correspond to any of these conditions. Although our patient had bilateral hemisphere injury, pseudobulbar palsy can be excluded as a possible cause for the frequent laughing in the present case because other pseudobulbar signs, such as dysarthria, dysphagia or increased jaw jerk and gag reflex, were not present and our patient's laughing occurred in the context of elevated mood.¹²

Secondary mania after focal brain injury has been reported in patients with lesions such as right temporal astrocytoma,¹³ right parietal chondroma¹⁴ and right thalamic infarction,¹⁵ as well as traumatic contusions in the right temporoparietal and bilateral orbitofrontal¹⁶ regions, suggesting that lesion localization and the aetiologies of the focal brain lesions causing secondary mania are diverse. Despite this diversity, there is a tendency for secondary mania to occur more commonly after right rather than left hemisphere injury.^{10,13–16} In addition, the right frontotemporal pathways connecting the orbitofrontal area with the basal temporal regions of the limbic system, when injured, are known to play a critical role in producing secondary mania.¹⁰ That is, the disruption of these frontotemporal pathways disconnects the prefrontal cortex from the limbic system, thereby releasing frontal inhibitions on the limbic system. The brain MRI in the present case showed diffuse bilateral subcortical and basal ganglia lesions. Thus, the frontal white matter lesion in the present case may have prevented frontal inhibition from being transmitted to the limbic system, resulting in mania that manifested as a burst of limbic activity.

Acknowledgement

This study was supported by a grant from the Korea Health 21 R&D Project, Ministry of Health & Welfare, Republic of Korea (A050079).

doi:10.1016/j.jocn.2005.10.013

References

1. Choi IS. Delayed neurologic sequelae in carbon monoxide intoxication. *Arch Neurol* 1983;**40**:433–43.
2. Min SK. A brain syndrome associated with delayed neuropsychiatric sequelae following acute carbon monoxide intoxication. *Acta Psychiatr Scand* 1986;**73**:80–6.
3. Mizutani T, Shiozawa R, Takemori S, et al. Delayed post anoxic encephalopathy without relation to carbon monoxide poisoning. *Intern Med* 1993;**32**:430–3.
4. Evans DL, Byerly MJ, Greer RA. Secondary mania: diagnosis and treatment. *J Clin Psychiatry* 1995;**56** (Suppl. 3):31–7.
5. Yagnik PM, Burns RA. Delayed anoxic encephalopathy presenting with psychiatric symptoms. *Psychosomatics* 1984;**25**:413–5.
6. Collins MN, Jacobson RR. Changing interactions between bipolar affective disorder and anoxic brain damage. *Br J Psychiatry* 1990;**156**:736–40.
7. Kumar A, Agarwal M. Secondary affective disorder in survivor of cardiac arrest. A case report. *Br J Psychiatry* 1988;**153**:836–9.
8. American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders. 4th edn. (DSM-IV). Washington DC: American Psychiatric Association; 1994.
9. Choi SH, Na DL, Kwon HM, et al. The Korean version of the neuropsychiatric inventory: a scoring tool for neuropsychiatric disturbance in dementia patients. *J Korean Med Sci* 2000;**15**:609–15.
10. Gafoor R, O'Keane V. Three case reports of secondary mania: evidence supporting a right frontotemporal locus. *Eur Psychiatry* 2003;**18**:32–3.
11. Krauthammer C, Klerman GL. Secondary mania: manic syndromes associated with antecedent physical illness or drugs. *Arch Gen Psychiatry* 1978;**35**:1333–9.
12. Flora GC. Pseudobulbar palsy. *Minn Med* 1970;**53**:569–72.
13. Sokolski KN, Denson TF. Exacerbation of mania secondary to right temporal lobe astrocytoma in a bipolar patient previously stabilized on valproate. *Cogn Behav Neurol* 2003;**16**:234–8.
14. Salazar-Calderon Perriggo VH, Oommen KJ, Sobonya RE. Silent solitary right parietal chondroma resulting in secondary mania. *Clin Neuropathol* 1993;**12**:325–9.
15. Kulisevsky J, Berthier ML, Pujol J. Hemiballismus and secondary mania following a right thalamic infarction. *Neurology* 1993;**43**:1422–4.
16. Bakchine S, Lacomblez L, Benoit N, et al. Manic-like state after bilateral orbitofrontal and right temporoparietal injury: efficacy of clonidine. *Neurology* 1989;**39**:777–81.

Acute sensory neuronopathy as the presenting symptom of Sjögren's syndrome

Nizar Souayah^{a,*}, Peter Siao Tick Chong^b, Didier Cros^b

^a Neurology Department, New Jersey Medical School, 90 Bergen Street, Suite 8100, Newark, New Jersey 07101, USA

^b Neurophysiology Lab, Neurology Department, Massachusetts General Hospital, Harvard Medical School, Boston, Massachusetts, USA

Received 7 October 2005; accepted 8 December 2005

* Corresponding author. Tel.: +1 973 972 8577; fax: +1 973 972 8738.
E-mail address: souayani@umdnj.edu (N. Souayah).