

Punding following posterior cerebral artery infarction: a case report and literature review

Cho-I Lin¹
 Yu-Ching Lin¹⁻³
 Wei-Chih Lien¹
 Pei-Chun Hsieh¹
 Sheng-Han Lin¹
 Ta-Shen Kuan^{1,2}

¹Department of Physical Medicine and Rehabilitation, National Cheng Kung University Hospital, College of Medicine, National Cheng Kung University, Tainan, Taiwan;

²Department of Physical Medicine and Rehabilitation, College of Medicine, National Cheng Kung University, Tainan, Taiwan; ³Medical Device Innovation Center, National Cheng Kung University, Tainan, Taiwan

Introduction: Punding is a complex stereotyped behavior, characterized by excessiveness, non-goal orientation, and repetitiveness. It is mostly associated with Parkinson's disease, and very few cases in non-Parkinson's disease have been reported. We report a case of punding associated with supratentorial ischemic stroke.

Case presentation: We present a 70-year-old man with left posterior cerebral artery infarction with quetiapine-induced punding manifesting as repetitive unidirectional body turning. Remission of punding behavior ensued after cessation of quetiapine and administration of clonazepam.

Conclusion: This case describes the clinical course of quetiapine-induced punding in a patient with left posterior cerebral artery infarction. It suggests clonazepam may serve as a treatment option for poststroke punding.

Keywords: punding, stereotyped behavior, stroke, quetiapine, clonazepam

Introduction

Punding is a complex, stereotyped, prolonged, and non-goal oriented behavior, originally proposed by Rylander in 1972 to illustrate a unique repetitive behavior in illicit drug users.¹ Although it was first noted in drug abusers,^{1,2} most cases of punding have been reported in patients with Parkinson's disease (PD).³ Only one case report delineated the development of punding in the subacute stage of pontine infarction.⁴ The spectrum of punding varies from as simple as hair brushing or paper shuffling, to more complicated activities such as hoarding, gardening, and tinkering with machinery.³ Cortico-striato-thalamo-cortical circuit has been postulated to be involved in the pathogenesis of punding, while its mechanism remains to be elucidated.³ Here we report a case of punding following left posterior cerebral artery infarction manifesting as repetitive unidirectional body turning. The condition persisted for 19 days and subsided after the administration of clonazepam.

Case presentation

A 70-year-old right-handed man was presented to our emergency room (ER) with acute onset of incoherent speech. His medical history was significant for hypertension and dyslipidemia even though he was not on antihypertensive, hypolipidemic nor other agents. The patient was a retired train operator and remained independent in activities of daily living until this episode. On examination, he was noted to have Wernicke's aphasia. There was no other cranial nerve palsy. Muscle strength of four extremities scored 5/5 according to the Medical Research Council (MRC) scale. Unenhanced computed tomography (CT) upon his arrival to the ER and magnetic resonance imaging (MRI) of the brain (Figure 1) 2 days later confirmed the diagnosis

Correspondence: Ta-Shen Kuan
 Department of Physical Medicine and Rehabilitation, National Cheng Kung University Hospital, College of Medicine, National Cheng Kung University, 138 Sheng-Li Road, Tainan 704, Taiwan
 Tel +886 6 235 3535 ext 5242
 Fax +886 6 276 6106
 Email kuan@mail.ncku.edu.tw

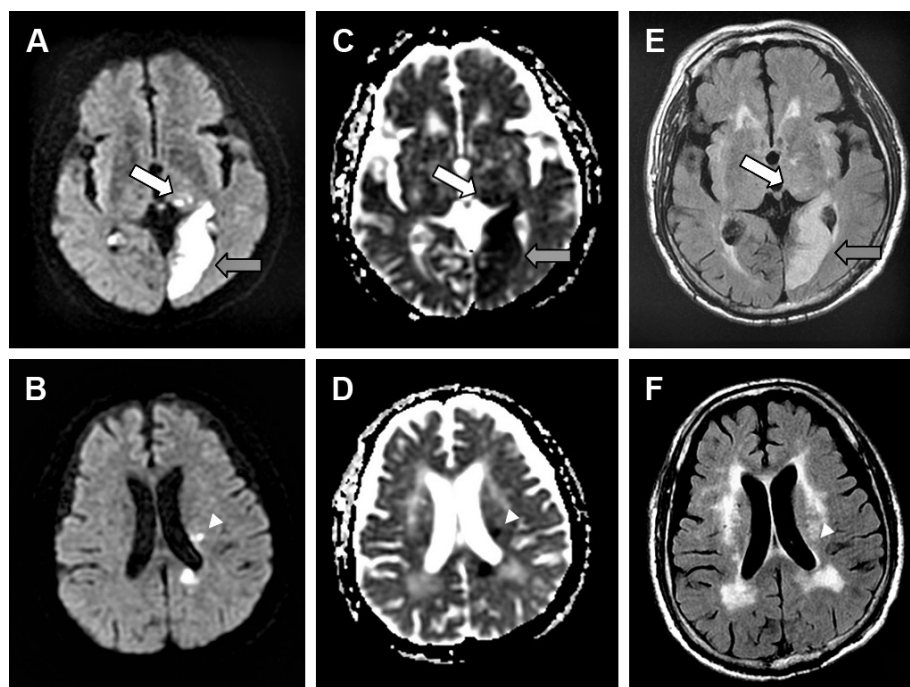


Figure 1 Magnetic resonance imaging results.

Notes: Hyperintensities noted in the left occipital lobe, corona radiata, and thalamus on transaxial diffusion weighted magnetic resonance imaging (**A, B**), and corresponding hypointensities on apparent diffusion coefficient map (**C, D**) suggested restricted diffusion resulting from an acute ischemic insult. Standard transaxial T2-weighted fluid attenuation inversion recovery sequence indicated hyperintensities in the corresponding region (**E, F**). Gray arrow, left occipital lobe; white arrow head, corona radiata; white arrow, thalamus.

of acute infarction in the territory of the left posterior cerebral artery, involving left occipito-temporo-parietal lobe, corona radiata, and thalamus. He was admitted to the neurology ward on the same day as the ER visit. His stroke deteriorated and he became hemiplegic on the right side with MRC score of 0/5 4 days after admission. His condition improved 2 days later, after hydration.

Ten days after the stroke, the patient was transferred to a rehabilitation ward. During an interview with a physiatrist, he was noted to have transcortical sensory aphasia and right-sided hemiparesis with MRC score of 3/5. Behavior during the initial phase of rehabilitation was characterized by agitation, reluctance, and occasional episodes of yelling. He started on quetiapine 50 mg at bedtime for intermittent irritability and poor sleep without concurrent administration of other hypnotics or dopaminergic medications. Eight days after initiation of quetiapine and 22 days after the stroke, he exerted unidirectional turning of his whole body to his right by 90° in a clockwise fashion repetitively. Such behavior could last 6–8 hours during daytime and the patient appeared to be preoccupied. It could be interrupted temporarily by gesture command, and would subside while he slept. He became furious when asked to stop his activities. Electroencephalography at that time indicated no epileptiform discharge. He started on oral clonazepam

(0.25 mg daily at first, gradually up-titrated to 0.5 mg twice daily within 1 week) 2 days after the onset of such behavior, and quetiapine was discontinued 2 days later. A gradual reduction in the duration of the stereotyped behavior was noted, and total remission was achieved 17 days after initiation of clonazepam therapy, and 15 days after cessation of quetiapine. The dosage of clonazepam was tapered off over a 1-week period (from 0.5 mg twice daily to 0.5 mg daily for the first 4 days, then 0.25 mg daily for another 3 days, and then discontinued). There was no recurrence of punding in the following 6 months of follow-up.

Written informed consent was obtained from the patient and his guardians for publication of the case details, and any accompanying images.

Discussion

Punding is a complex stereotyped behavior featured by intense fascination with repetitive, excessive, non-goal oriented behavior.^{3,4} It is usually executed at the expense of other motor activities.^{3,5,6} The essential elements in punding include excessiveness, disruptiveness, and interruption resulting in anger.^{3,4} Patients with punding tend to be preoccupied by their chosen activities and withdraw into themselves, and this interferes with their interactions with the outer world.^{3–5} The above-mentioned characteristics of punding were all present

in our case. The behavior pattern in our patient manifested as repetitive unidirectional body turning to the paretic side. Another possible explanation for such phenomenon was clonic perseveration, in which the inappropriate repetition of an action persisted despite absence of an ongoing cue.⁷ However, the phenomenon observed in our case was not triggered by external cues, and that made clonic perseveration a less likely explanation.

Most cases of punning have been reported in patients with PD.³ Observation of patients with PD suggests the spectrum of behavior pattern is influenced by gender, occupation, or even hobbies.³ However, such associations are not noted in our case. The behavior pattern of punning ranges from simple form such as repetitive hair brushing and repeatedly opening and closing handbags, to complex form like gardening.³ There is only one case report in the literature describing the emergence of punning following brainstem infarction.⁴ Moreover, very few cases of stroke-induced stereotyped behavior have been reported. Available PubMed literature retrieved on January 19, 2017 by searching for “stereotypy”, “stereotyped behavior”, and “stroke” has been summarized in Table 1, and additional relevant case reports referenced by these publications are also included. Our review suggests basal ganglion and thalamus (two out of seven cases, respectively) are the most frequently involved locations, and the others include pons, cerebellum, frontal and parietal lobes. The mean latency from onset of cerebrovascular events to the occurrence of stereotyped behavior is 38.6 days (ranging from 5 to 90 days). In our case, the acute infarction in the territory of left posterior cerebral artery mainly involved the left occipital lobe and left thalamus. The occipital lobe is known for vision processing but has not been reported to be associated with stereotyped behavior. In contrast, there were two case reports signifying the involvement of thalamus in the development of poststroke stereotyped behavior.^{8,9}

Although the underlying pathophysiology of punning remains uncertain, it has been attributed to excessive dopaminergic stimulation.³ Indeed, most punning behavior is observed in levodopa-treated PD patients, and reduction of the dose of antiparkinsonian medications results in improvement in punning behavior.³ Other medications, aside from dopaminergic replacement therapy, may play a relevant role in development of punning.^{3,4,10} Miwa et al reported emergence of punning after starting quetiapine therapy in two PD patients, the dosing of quetiapine ranged from 100–200 mg per day.¹⁰ Quetiapine is an atypical antipsychotic drug that interacts with multiple

neurotransmitter receptors, with a higher affinity for serotonin receptors (5-hydroxytryptamine 2, 5-HT₂) than its dopamine counterpart (D1 or D2).^{3,10} It has been shown to induce dopamine release by serotonin 5-HT₂ receptor blockade.³ In our case, although the dose of quetiapine was relatively low, the temporal relationship of quetiapine initiation and punning occurrence, as well as its cessation and subsidence of punning supports a reciprocal link between quetiapine and punning. Furthermore, as infarction in posterior cerebral artery territory, especially the involvement of thalamus, disrupted the cortico-striato-thalamo-cortical pathway, the administration of quetiapine might exert a synergistic effect and both play pivotal roles in the development of punning in our case.

Cortico-striato-thalamo-cortical circuits that originate in the prefrontal cortex and project to the striatum and thalamus, and then return to prefrontal cortex, have been proven to play a vital role in the pathogenesis of behavioral and movement disorders.^{8,9,11} Theoretically, reduced basal ganglia output would result in disinhibition of the cortico-striato-thalamo-cortical circuits, exciting cortical motor areas to exhibit movements that ought to be suppressed.^{6,12} Gamma-aminobutyric acid (GABA) has been shown to be a vital neurotransmitter in the basal ganglia output pathway.^{12,13} Clonazepam is a benzodiazepine and acts as an enhancer of GABA-mediated effects.^{12,13} As a consequence, it might increase GABAergic neurotransmission and thereby modulate movement disorders.^{6,12} In clinical practice, clonazepam has been proven to be effective in several poststroke movement disorders including hemichorea, hemiballism, dystonia, myoclonus, and tremor.^{13,14}

To the best of our knowledge, complex stereotyped behavior resulting from cerebrovascular accidents is very rare in the existing literature. We reported the first case depicting the occurrence of punning following supratentorial ischemic stroke. Punning greatly impeded rehabilitation participation and endangered social interaction in our case, and that is a common issue among other patients with punning.^{3,10,15,16} Considering the wide application of clonazepam and its proven effectiveness in poststroke movement disorder,^{12,13} we treated this patient with clonazepam dosed as 0.5 mg twice a day. Punning remitted thoroughly after 17 days of clonazepam therapy, and there was no recurrence of punning after discontinuation of clonazepam in the following 6 months. The therapeutic effect of clonazepam appeared to be optimal in our patient. It warrants further investigation to provide more evidence of clonazepam for treatment of poststroke punning.

Table 1 Literature review of punning and stereotyped behavior following cerebrovascular accidents

References	Age	Gender	Handedness	Occupation	Comorbidities	Latency from onset (days)	Behavior pattern	Etiology	Anatomical locations	Treatment	Outcome
Nguyen et al ⁴	54	M	R	n/a	HTN hypercholesterolemia, TIA, depression, tobacco and alcohol abuse	n/a	Food purchasing and hoarding; writing, copying, and organizing of recipes	Ischemic stroke	Right ventral pons	Sertraline 100–150 mg for 2 weeks	Complete remission
Arauco et al ⁸	22	F	n/a	n/a	Nil	90	Repetitive hand flapping, finger snapping close to the ear, alternating from right to left, ordering, and touching compulsions	Spontaneous subarachnoid hemorrhage	Bilateral thalami and frontal lobes	n/a	n/a
Pandey and sarma ⁹	40	M	n/a	n/a	Old CVA with left hemiparesis	5	Purposeless repetitive right hand tapping	Ischemic stroke	Left thalamus	n/a	Complete remission
McGrath et al ⁵	21	M	n/a	College student	Recurrent CVAs with encephalomalacia	70	Episodic agitation, yelling, and repetitive body rocking	Subdural hematoma	Left frontal and parietal lobes	Sertraline 50–100 mg for 1 month	Complete remission
Maraganore et al ¹⁶	17	M	n/a	n/a	Aortic coarctation	n/a	Circling gait-interrupting behavior and complex stereotyped movements of hands	Ischemic stroke	Right putamen	n/a	n/a
Kulisevsky et al ¹⁷	69	M	R	n/a	HTN, DM	21	Opening and closing movements of the mouth, body rocking while seated; floor pacing, and marching in place while standing	Ischemic stroke	Right lentiform nucleus	Pimozide (1 mg/day) for 6 weeks	Complete remission of restless motor movements
Lee et al ¹⁸	71	F	n/a	n/a	Hypothyroidism	7	Repetitive supination and pronation of right hand or thigh rubbing when seated; flexion and extension of the right elbow while walking	Ischemic stroke	Right cerebellum and left lingual gyrus	Cilostazol, donepezil	Complete remission

Abbreviations: CVAs, cerebrovascular accidents; HTN, hypertension; DM, diabetes mellitus; TIA, transient ischemic attack; n/a, not available; M, male; F, female; R, right.

Conclusion

This case report presented the clinical course of quetiapine-induced punning in a patient with left posterior cerebral artery infarction. We also described the effectiveness of clonazepam for punning. Although there is no standard treatment, clonazepam may be a treatment option for poststroke punning.

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Disclosure

The authors report no conflicts of interest in this work.

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