

## LETTER TO THE EDITOR

**Amelioration of pathological yawning after tracheostomy in a patient with locked-in syndrome**C.-C. Chang<sup>a</sup>, S.-T. Chang<sup>a</sup>,  
H.-Y. Chang<sup>b</sup> and K.-C. Tsai<sup>a</sup><sup>a</sup>Department of Physical Medicine and Rehabilitation, Tri-Service General Hospital, School of Medicine, National Defense Medical Center, Taipei, Taiwan; and <sup>b</sup>Department of Nursing, Tri-Service General Hospital, National Defense Medical Center, Taipei, Taiwan**OnlineOnly:** This article is available online only at [www.blackwell-synergy.com](http://www.blackwell-synergy.com)Correspondence: Shin-Tsu Chang, Department of Physical Medicine and Rehabilitation, Tri-Service General Hospital, National Defense Medical Center, No. 325, Section 2, Cheng-Kung Road, Neihu District, Taipei 114, Taiwan (tel.: +886 2 8792 3311; fax: +886 2 8792 7162; e-mail: [stchang@ms87.url.com.tw](mailto:stchang@ms87.url.com.tw)).**Keywords:** dead space, locked-in syndrome, tracheostomy, vagus nerve, yawning

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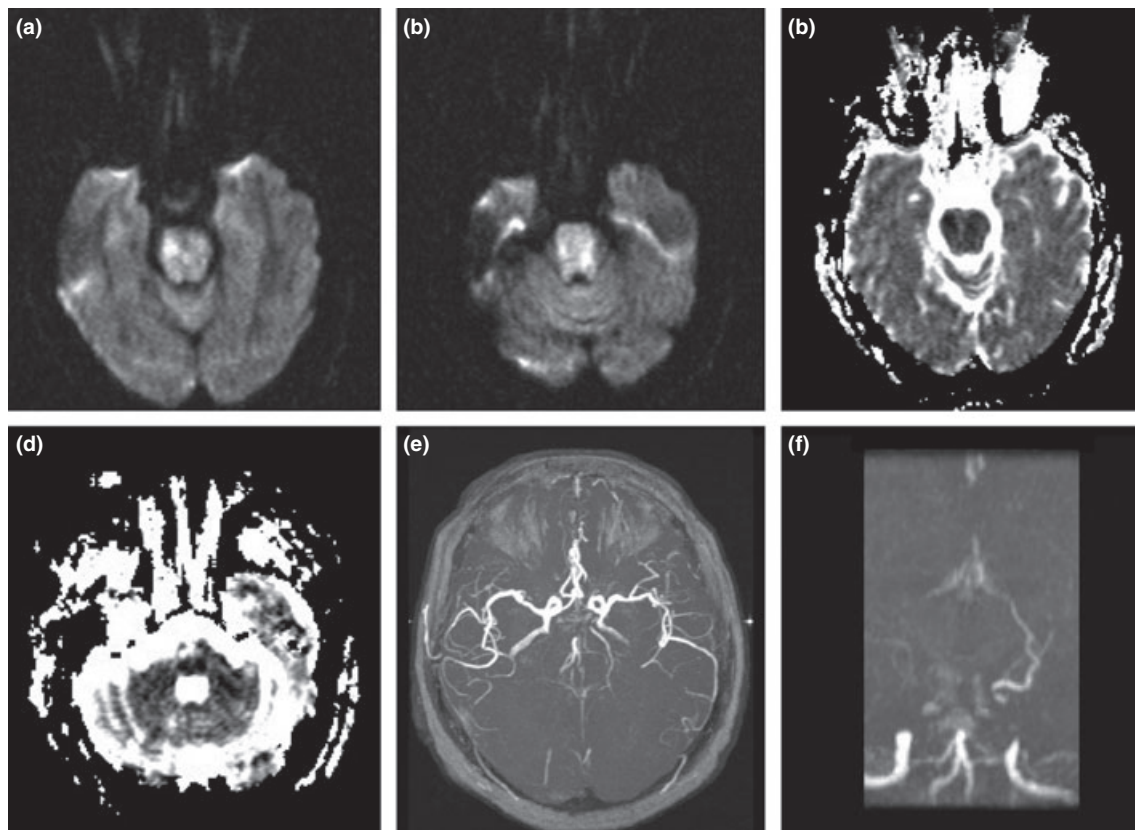
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**Case report**

A 66-year-old female with a past history of hypertension, who initially presented with four limbs weakness for a duration of 3 h, poor visual acuity, ataxia and slurred speech at ER and progressed to drowsiness, tetraplegia with episodes of unjustified excessive and repetitive yawning and acute respiratory failure within few hours. On admission, the patient was drowsy and showed a positive doll's eye sign and positive gag reflex. Her uvula and tongue deviated to the left and weakness of facial muscle caused drooling. The right upper and lower extremity strength were rated 3+/5 and 2+/5 separately. The left upper and lower extremity strength were rated

2+/5 and 0+/5 respectively. Deep tendon reflexes were enhanced and bilateral Babinski signs were present. A brain magnetic resonance imaging (MRI) scan showed an infarct in most parts of the pons and another small lesion in right medial occipital lobe appearing as increased signal intensity on DWI with corresponding low ADC. MRI showed total occlusion of the middle and distal thirds of basilar artery. Besides, the left posterior cerebral artery arose from the left internal carotid artery with focal stenosis at its proximal portion and there was an aneurysm about 3 mm in size located at the anterior communicating artery (Fig. 1).

The patient developed acute respiratory failure and progress to flaccid tetraplegia several hours after admission. The frequency of excessive yawning persisted despite adequate sleep at night. An endotracheal tube was inserted as a prerequisite of subsequent ventilatory support. Several days later, she still had limitations



**Figure 1** On admission, brain magnetic resonance imaging (MRI) showing an infarct in most parts of the pons on DWI (a, b) with corresponding low ADC (c, d). Brain MRI showing total occlusion of the middle and distal thirds of basilar artery (e, f). The left posterior cerebral artery arising from the left internal carotid artery with focal stenosis at its proximal portion and an aneurysm about 3 mm in size located at the anterior communicating artery were found (e).

with regards to eyeball movement, prominent hypomimia, bilateral masticatory spasms and persistent trismus and more frequent yawning with an extreme opening of the mouth. The patient developed anarthria and dysphagia and had soft palate paresis, and brisk gag reflexes. Ten days after admission, she received tracheostomy under local anaesthesia at bedside in the intensive care unit. After the tracheostomy tube was secured, the patient's breathing tube was removed. She made an uneventful recovery after the procedure. To our surprise, the frequency of yawning gradually decreased and returned to normal within hours after the tracheostomy. The duration of excessive pathological yawning persisted only for 10 days.

### Discussion

The precise role of yawning in human physiology remains unclear, but the central neural structures that control yawning are presumably located in the brain stem close to or within other respiratory and vasomotor centres [1]. The earliest appearance of yawning was observed in a 15-week-old embryo [2]. Frequency of yawning decreases with brain maturation when the connections between the limbic system and the forebrain areas are already established [3]. Besides, another factor contributing to yawning is total dead space of the respiratory system which would be relatively larger in paediatric subjects [4]. Because it is interesting to note that the frequency of yawning gradually decreased and returned to normal within hours after the tracheostomy was performed in our patient presented with locked-in syndrome, we hypothesized that initial enlarged dead space after endo-

tracheal tube intubation would be another trigger of yawning because our patient's dead space was reduced by tracheostomy which may be one of the major factors capable of terminating the pathological yawning.

Others postulated that repetitive yawning in cardiac tamponade might be secondary to phrenic nerve irritation [5,6]. Another hypothesis is that yawning stimulates a haemodynamic compensatory response to tamponade [7]. However, there was no tamponade found in our patient. However, there was possibility that the phrenic nerve of the patient might be influenced by vagus nerve manipulation or stimulation during the tracheostomy similar to the reports proposed by Eldridge and Millhorn who demonstrated that 0.5 min duration of vagus nerve stimulation induces a reduction (< 1 min duration) in phrenic amplitude and frequency after stimulation [8]. Besides, Zhang *et al.* [9] suggested that a relatively longer post-stimulation inhibitory memory is possible if using episodic and longer vagus nerve stimulation. For many years, episodic vagus nerve stimulation has been applied clinically as a treatment for patients with refractory epilepsy [11]. However, the mechanisms and the consequence of long-term vagus nerve stimulation remain unclear [10]. We postulate that perioperative vagus nerve manipulation or stimulation might occur during the tracheostomy, which probably played a role in the triggering or even termination of pathological yawning because there have been similar treatments using tracheostomy and cauterization applied in the treatment of epilepsy in the early 19th century [11].

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