European Journal of Neurology 2007, **14:** 461–463 doi:10.1111/j.1468-1331.2006.01645.x

SHORT COMMUNICATION

Paroxysmal sneezing at the onset of lateral medullary syndrome: cause or consequence?

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Keywords:
lateral medullary syndrome, sneezing, Wallenberg's syndrome

Received 15 May 2006
Accepted 18 August 2006

Sneezing is known to precede lateral medullary syndrome (LMS). It is usually interpreted as the precipitating cause for a vertebral artery dissection that subsequently causes LMS. Through two case reports and a literature review, we aim to challenge the concept that sneezing at the onset of LMS implies that a dissection is the underlying cause. An 82-year-old man and a 54-year-old man both reported unprovoked explosive pathological sneezing at the onset of the LMS without any delay between sneezing and the other LMS symptoms. Both denied neck trauma or neck pain. There was no conclusive evidence for vertebral artery dissection in either case. Paroxysmal sneezing can be an initial manifestation of lateral medullary ischemia and may not necessarily indicate an underlying vertebral artery dissection as the cause.

The lateral medullary syndrome (LMS), or Wallenberg's syndrome, often results from occlusion or dissection of the vertebral artery. Vertebral artery dissection has been blamed on many different life events, such as sneezing [1]. Paroxysmal sneezing at the onset of LMS is usually interpreted as a cause, since a violent sneeze could potentially result in a vertebral artery dissection causing LMS [2]. In this study, we describe two cases where paroxysmal sneezing is likely to be an initial symptom of LMS rather than the precipitating cause.

Report of two cases

Case one

A hypertensive 82-year-old man experienced a sudden attack of violent sneezing while resting in a recliner. Sneezing was unusual for him, and the intensity and duration of the sneezes seemed odd. Immediately after this atypical sneezing attack subsided, he noticed severe vertigo and fell to the right. He denied neck pain and had no prior neck trauma or manipulation. Upon neurological examination, he was found to have a hoarse voice, right ptosis and miosis, right-sided dysmetria, and truncal ataxia. Head computed tomography was unremarkable except for old periventricular lacunar infarctions. Due to metal in the orbit, magnetic resonance imaging and magnetic resonance angiography could not be obtained. Computed tomography angiography of head and neck showed intact vessels with no evidence of vertebral artery dissection (Fig. 1). He did not have any further sneezing episodes during his hospitalization or report inability to complete a sneeze.

Case two

A 54-year-old man with hypertension, hyperlipidemia, and coronary artery disease experienced an abrupt onset of 15 violent sneezes while watching television. This had never happened to him before. Once the sneezing attack ceased, he noticed that the right side of his face was numb. He listed to the right while walking. His voice was hoarse. He denied neck pain and had no prior neck trauma or manipulation. Examination revealed right miosis and ptosis, right horizontal and torsional nystagmus, and right-sided dysmetria. He had a wide-based gait and veered to the right. Magnetic resonance imaging of the brain showed an acute right lateral medullary infarct in the rostral medulla (Fig. 2). Magnetic resonance angiography of the neck showed absent or reduced flow of the right vertebral artery and was inconclusive for either a dissection or atherosclerosis (Fig. 3). He never lost the ability to sneeze and had no further explosive sneezing attacks.

Discussion

Sneezing is a complex protective respiratory reflex that has two phases: nasal and respiratory. The nasal phase is mediated by trigeminal afferents feeding back to the presumed medullary sneeze center. The respiratory

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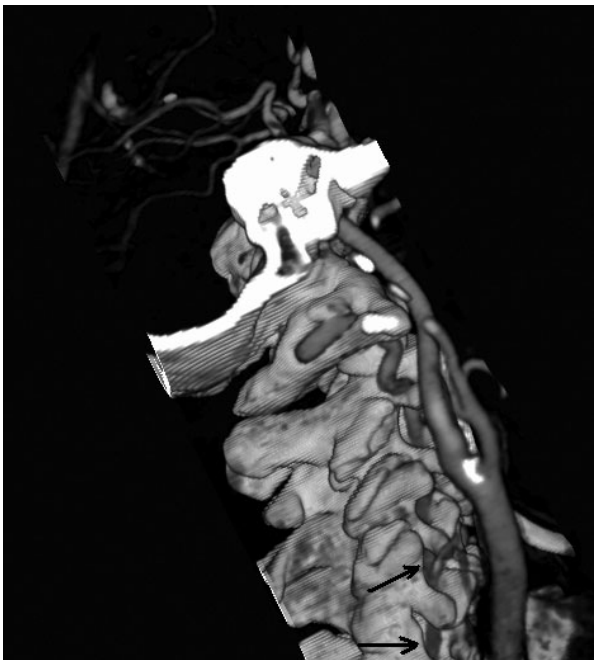

Figure 1 Computed tomography angiogram of the neck in case one, lateral view. Right carotid artery is visualized with an intact right vertebral artery (arrows).

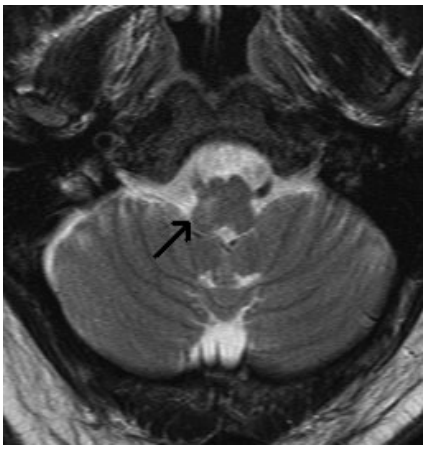

Figure 2 Magnetic resonance imaging-T2 weighted image in case two. Axial section through the level of the rostral medulla showing a right lateral hyperintense signal (arrow).

phase commences when a critical number of inspiratory and expiratory neurons are recruited by the sneeze center. Recruitment of these neurons increases activity in the vagus, phrenic, and intercostal nerves to the appropriate musculature. Manifestations of this phase consist of the following sequence of events: eye closure, deep inspiration, pharyngeal closure, forceful expiration, dilatation of the glottis, explosive air release through the mouth and nose, and expulsion of mucus and irritants [3].

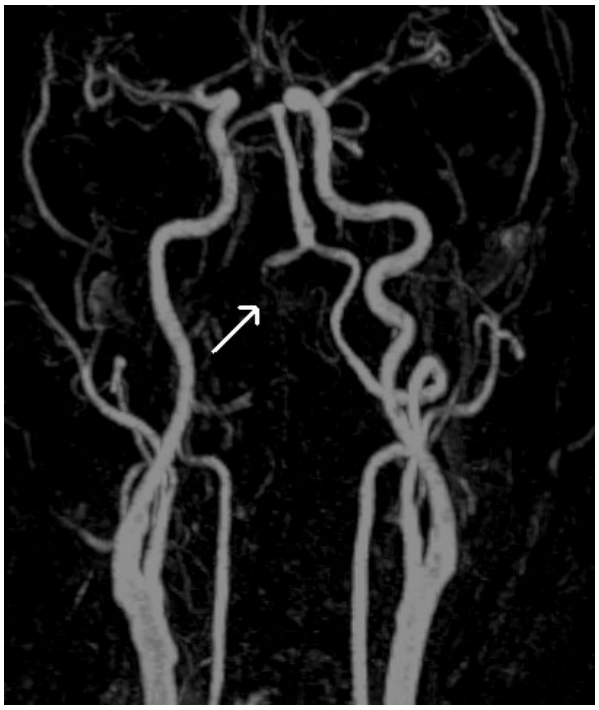

Figure 3 Post-contrast magnetic resonance angiogram of the neck in case two. Coronal view reveals an absence or reduction of flow consistent with a high grade stenosis/occlusion of the right vertebral artery.

Nonaka *et al.* used electrical microstimulation in precollicular-postmammillary decerebrate cats to localize the sneezing center to the medulla, at the ventromedial border of the descending tract and spinal nucleus of the trigeminal nerve and the immediately subjacent reticular formation [4]. Not surprisingly, the loss of the ability to sneeze has been reported in LMS [5]. Further localization of the human sneeze center was described by Seijo *et al.* in a patient with right LMS, initially presenting with violent sneezes and followed by brief loss of the sneeze reflex with eventual recovery. The lesion was located proximal to the interpolaris-caudalis area of the right trigeminal spinal tract and nucleus [6]. In LMS, hiccups tend to occur mainly with lesions in the dorsolateral region of the middle medulla [7] and swallowing abnormalities occurred with lesions of the rostral part of the dorsal medulla [8]. Neither of our patients complained of hiccups. Dysphagia was not reported on admission and formal swallowing studies performed within 48 h were unremarkable for both patients.

Based on the compelling clinical features of our two cases, we hypothesize that paroxysmal sneezing at the onset of LMS can be a symptom of early ischemia in the medullary sneezing center. Other positive phenomena have been associated with brainstem ischemia. For

example, hiccups can occur at the onset of LMS [3]. Ropper reported abnormal movements during pontine infarctions [9]. We realize that it is often difficult to determine the temporal sequence of events in this setting. Bernat and Seijo each described a patient with LMS heralded by sneezes and wondered if these symptoms were a cause or a consequence of the stroke [2,6]. We base our hypothesis on the unusual characteristics of the sneezing, the timing with the LMS symptoms, and the absence of evidence to support dissection as a mechanism.

These sneezes were clearly pathological, very intense and atypical for the subjects. In addition, patients that develop vertebrobasilar ischemia after neck manipulation often report a variable delay between the exposure and the onset of symptoms [10]. In our cases, there was no delay between sneezing and the other brainstem symptoms, as it would be expected if the symptoms were caused by a secondary thrombus over a dissected artery. Furthermore, these patients did not have evidence of arterial dissection. The neuro-imaging studies showed intact vessels in the first case and inconclusive findings in the second case. Both patients had risk factors for atherosclerosis and denied having head or neck pain, a hallmark of a dissected vertebral artery [1]. The clinical and radiological features of all these cases suggest that paroxysmal sneezing can be an initial manifestation of LMS. Therefore, the presence of sneezing at the onset of LMS does not necessarily imply that dissection is the underlying mechanism.

Disclosure

No conflicts of interest to disclose.

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