



Insular seizures causing sleep-related breathlessness

Alexander D Nesbitt, Christopher A Kosky, Guy D Leschziner

Lancet 2013; 382: 1756

Sleep Disorders Centre, Guy's and St Thomas' NHS Foundation Trust, London, UK
 (A D Nesbitt MRCP, C A Kosky FRACP, G D Leschziner MRCP)

Correspondence to: Dr Guy Leschziner, Consultant Neurologist, Sleep Disorders Centre, Nuffield House, Guy's Hospital, Great Maze Pond, London SE1 9RT, UK
 guy.leschziner@gstt.nhs.uk

A 52-year-old woman was referred for assessment for sleep apnoea in October, 2010, with excessive daytime sleepiness, loud snoring, and regular waking from sleep with choking and breathlessness. She described regular events—occurring shortly after falling asleep—whereby she would suddenly wake up with a feeling of choking or being strangled for several seconds, accompanied by a sharp, central chest pain. These events were always associated with a feeling of dyspnoea and anxiety, lasting 1–2 min. Rarely she would feel a stiffening sensation of her right leg as the event subsided. After prompting, she recalled that with more severe events, she may have bitten the lateral margins of her tongue. She remained fully aware of her surroundings throughout, and was able to return to sleep afterwards. These events would happen once, and at most twice, during the night, 5 or 6 nights a week, and would worsen in severity during the late luteal phase of her menstrual cycle. She remembered having very similar, less frequent, and less severe events from the age of 11 years, which had been treated as panic attacks with intermittent oral chlorpromazine. Her birth, childhood, and developmental history were unremarkable. She had a background of mild asthma. There was no family history of epilepsy. Physical examination was normal. Standard overnight polysomnography demonstrated fragmented sleep and an apnoea-hypopnoea index of 5.4/h. An MRI scan of the brain was normal. A further full-montage overnight sleep EEG, and separate sleep-deprived wake EEG, showed brief focal bursts of irregular theta activity over the left temporal area during frequent awakenings from stage N2 sleep. An FDG-PET scan of the brain showed an asymmetrical hypometabolic focus in the region of the left insula extending into the left temporal lobe (figure). A diagnosis

of sleep-related focal epilepsy, localised to the left insular cortex, was made. Initiation of oral levetiracetam, escalated to a dose of 1500 mg nightly, resulted in suppression of almost all the patient's seizures, with great improvement in sleep quality and daytime functioning. At last follow-up in September, 2013, she was experiencing only very occasional attacks of much reduced severity, which would occur only in the few days preceding her menses.

The insular cortex lies deeply folded within the lateral sulcus of each hemisphere, covered by overlying areas of frontal, temporal, and parietal lobes. Penfield and Jasper¹ were the first to report the insular cortex's prominent roles in viscerosensory, visceromotor, and autonomic control, as well as somatosensory, pain, auditory, and speech processing. With the location of the insula and its functional connectivity with the central autonomic network, seizures arising from it can mimic temporal and frontal seizures,² and may more rarely cause ictal arrhythmias, including asystole.³ Scalp EEG may show interictal discharges of value in lateralising the side of the epileptic focus, but is otherwise unhelpful at differentiating seizures of insular origin from those originating from overlying areas of cortex.² Isnard and coworkers⁴ directly recorded insular seizures using implanted cortical EEG in six patients, and described key characteristics, thought to be highly specific for these attacks. All patients maintained full consciousness and immediately noted laryngeal discomfort as a choking or strangling sensation that was marked by anxiety and subjective dyspnoea. Perioral or truncal paraesthesiae were described, together with dysphonia and mutism in some patients, lasting up to a minute. The attacks typically ended with a stiffening sensation in a limb contralateral to the focus.⁴ Nocturnal choking and dyspnoea are frequently described symptoms of common disorders, such as obstructive sleep apnoea, heart failure, gastro-oesophageal reflux, laryngospasm, and panic disorder.⁵ Careful appraisal of additional symptoms suggestive of seizure activity should be done during the assessment of such patients.

Contributors

The patient was initially assessed by CAK and referred to GDL for specialist investigation and long-term management. ADN reviewed the case notes, wrote the initial draft and did the literature search with CAK and GDL. All edited and approved the final manuscript. Written consent to publish was obtained.

References

- 1 Penfield WG, Jasper HH. *Epilepsy and the functional anatomy of the human brain*. Boston: Little, Brown and Company, 1954.
- 2 Kriegel MF, Roberts DW, Jobst BC. Orbitofrontal and insular epilepsy. *J Clin Neurophysiol* 2012; 29: 385–91.
- 3 Sevcencu C, Struijk JJ. Autonomic alterations and cardiac changes in epilepsy. *Epilepsia* 2010; 51: 725–32.
- 4 Isnard J, Guenot M, Sindou M, Manguiere F. Clinical manifestations of insular lobe seizures: a stereo-electroencephalographic study. *Epilepsia* 2004; 45: 1079–90.
- 5 Davis KA, Cantor C, Maus D, Herman ST. A neurological cause of recurrent choking during sleep. *J Clin Sleep Med* 2008; 4: 586–87.

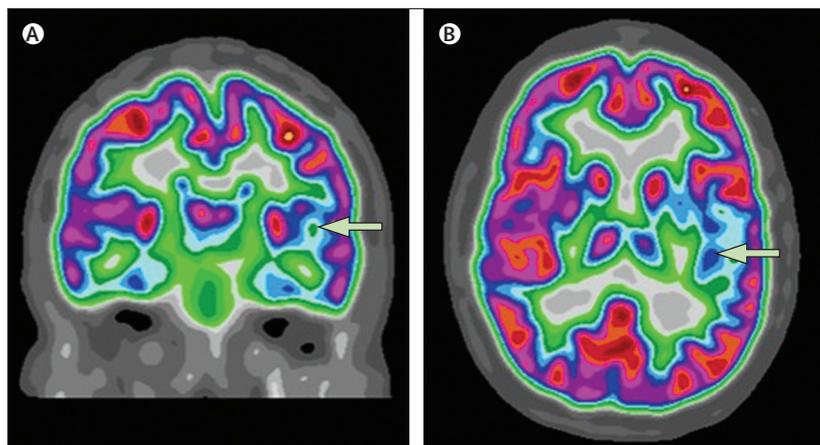


Figure: Insular seizures: FDG-PET scan

FDG-PET scan with low resolution computerised tomography showing hemispheric asymmetry with a hypometabolic focus in the region of the left insula, extending into the left temporal lobe (arrows) in both coronal (A) and transverse (B) planes.