Six months later, she had her sixth exacerbation that was characterised by inability to urinate, truncal and lower extremity numbness and quadriparesis. Physical examination at the time of this attack revealed monoplegia affecting the right leg with less than antigravity strength in the other three extremities. She had hyperreflexia in the upper extremities, clonus of the ankles and extensor plantar responses bilaterally. Visually evoked potentials showed a slight delay at 129 ms in the left eye. NMO IgG was negative. MRI of the brain and spinal cord during the sixth attack showed T2 and STIR abnormality from C4–C6 and T11–L1 within the central region of the spinal cord with no gadolinium enhancement. CSF was normal with no oligoclonal bands and normal IgG index.

We saw the patient 4 months after her sixth exacerbation at which time she reported a past history of pleuritis, erythema nodosum, oral and genital ulcers, malar rash, pathergy response and livedo reticularis. Rheumatological panel including ANA, SSA and SSB was negative. ACE levels and HTLV-I/II were normal. Vitamin B12, folate, lyme, RPR, TSH and thyroxine levels were negative or normal. Lupus anticoagulant IgM and IgG were initially positive at this point and were indeterminate (borderline) when checked 4 months later. Acetylcholine receptor antibody test was negative. She never had clinical evidence of MG or peripheral neuropathy and therefore further serological testing and nerve conduction tests were not indicated. A chest CT scan to look for evidence of sarcoidosis or pleuritis revealed a thymic mass.

The patient underwent thymectomy to remove the mass, determined histologically as thymic follicular hyperplasia. Following thymectomy her symptoms resolved completely. Six months after the surgery, methotrexate was discontinued. Except for mild fatigue after exertion and occasional mild burning and numbness in the right arm, she has been symptom free for an additional 23 months.

DISCUSSION

This patient was appropriately diagnosed with recurrent TM associated with Behcet's disease.[11] She did not meet criteria for systemic lupus erythematosis[12] or NMO.[13]

The patient has continued to have features of Behcet's syndrome with cutaneous and genital manifestations following thymectomy. These manifestations have decreased in frequency, recurring about 10 times a year prior to thymectomy, to 5–7 times a year following thymectomy. It is possible that the decreased frequency in recurrence of Behcet's may be partially attributed to resection of her thymus gland. However, remission of TM with persistence of Behcet's makes thymectomy more likely to have induced remission of TM in this patient.

This unusual case of TM and thymic follicular hyperplasia with recovery of complete neurological function after thymectomy suggests a possible causal relationship with TM. More research is needed to elucidate this association. Cases of TM should be investigated for thymic masses.

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Trouble at dinner: an unusual case of eating-induced seizures

Reflex seizures induced by eating are thought to be rare except in specific regions of Sri Lanka and India. They are predominantly of complex or simple partial type and are invariably associated with a symptomatic localisation-related epilepsy.[1]

Here, we report a patient presenting with unusual prandial-reflex-type ictal phenomenology and infratentorial, macrocystic lesion occupying the lower dorsal half of the medulla oblongata, anatomically co-localising with the dorsal vagal nucleus (DVN) and the nucleus tractus solitarius (NTS), diagnosed as haemangioblastoma on MRI.

CASE REPORT

A retired 67-year-old ex-lorry-driver presented with stereotyped attacks during meals. They were characterised by an odd metallic taste and a strange non-specific feeling after the first or second bite of the meal, without impairment of consciousness. After 2 years, these evolved into more serious attacks in which he would cough for about 30 seconds and then vomit. Following this, he was amnesic and confused for up to 30 minutes. During this phase, he was able to talk but mainly repeated simple questions and there were no other physical manifestations. The attacks appeared infrequently, up to several times per month, and were always independent of the taste, quality or other physical characteristics of the ingested food. On one separate occasion, he experienced an ill-defined tingling sensation on his left leg, which ascended over a few seconds to his left arm and lasted for a few minutes. He was started on carbamazepine 400 mg and aspirin 75 mg daily and, within 6 days, these episodes stopped and he has remained seizure-free for 8 months since. Four years ago, he had been successfully treated for non-Hodgkin's lymphoma with chemotherapy and had received radiotherapy for prostate cancer in 2001. He was otherwise healthy. A general physical examination was normal. An MRI brain scan showed a cystic lesion with nodular enhancement in the dorsal medulla, which was diagnosed as a haemangioblastoma on radiological grounds; no other abnormality was shown (fig 1). A waking, interictal EEG was normal.

DISCUSSION

Phenomena of reflex epileptogenesis is still poorly understood and, consequently, reports on patients with complex reflex seizures continue to yield considerable interest for epileptologists. Accounts of seizures with eating are relatively rare and are thought to be associated with localised or regional epileptiform activity either from temporolimbic structures or extralimbic regions. The latter subtype was previously reported in connection with more obvious suprasylvian structural lesions, including the operculum and the insula cortices, and was proposed to involve possible activation by thalamocortical afferents.[1]

Recently, a model by which the triggers for reflex seizures interact with pools of susceptible neurons to generate epileptogenic brain activity or clinical seizures in susceptible individuals was proposed.[1] In eating epilepsy, the specific seizure-triggering mechanisms are uncertain and, in the past, the sensation of smell, taste, gastric

distension, hyperglycaemia, the masticatory movements of the jaw, tongue action and swallowing were all proposed as contributing factors.[1] It is unclear which specific triggering factors were crucial in our patient, but they are likely to have included chewing and swallowing of the initial mouthful.

In the recent functional MRI imaging studies, the insula and frontoparietal operculum area were selectively shown to be activated by swallowing.[2] Previous studies and clinical data implicated those same areas in gustatory aura. Furthermore, intracranially recorded seizures involving the insula are associated with a characteristic sequence of laryngeal discomfort, viscerosensitive manifestations and oroalimentary phenomena (chewing, swallowing, dysarthria) with apparently preserved consciousness.[3] All three appear to be of relevance in our patient. Our case in other aspects resembles that described by Rosseti *et al.* in which the insular seizures occurred exclusively with simple partial gustatory and brachiocrural somatosensory phenomena whilst lacking the surface ictal EEG correlate.[3]

A causal connection between a structural lesion and prandial epilepsy is seldom demonstrated.[1] O'Brien and his colleagues proposed that lesions in those patients interfered with the inhibition or processing of sensory inputs to the cortex and, consequently, led to excitation and production of a critical mass of synchronised cortical neurons.[4] Although traditionally controversial, seizures of
subcortical and infratentorial origin have been demonstrated in animal models and are well documented in patients with hypothalamic hamartomas and cerebellar dysplastic lesions.[5 6] To our knowledge, this is the first report to show the coexistance of eating seizures and an infratentorial lesion. We cannot exclude the possibility that this lesion is coincidental and that the patient also has cryptogenic insular epilepsy. However, we hypothesise that his eating attacks result from interference with the processing of sensory inputs to the cortex. The lesion co-localises with medullary structures, including the DVN and NTS. The DVN relays general visceral afferents and the NTS gustatory afferents, which were both intimately involved in his episodes. Furthermore, they are known to project via parabrachial and thalamic nuclei to the insula and other surrounding temporolimbic cortical regions, all of which are commonly associated with gustatory aura and epileptiform activity in association with eating seizures.[1] Finally, the growing body of clinical and research data show that the electrical stimulation of the vagal nerve can modify the electrophysiological profile of cortical neurons and, as such, probably presents one of the pathophysiological mechanisms behind the anti-epileptogenic effect of vagus nerve stimulation treatment in focal-onset seizures.[7]

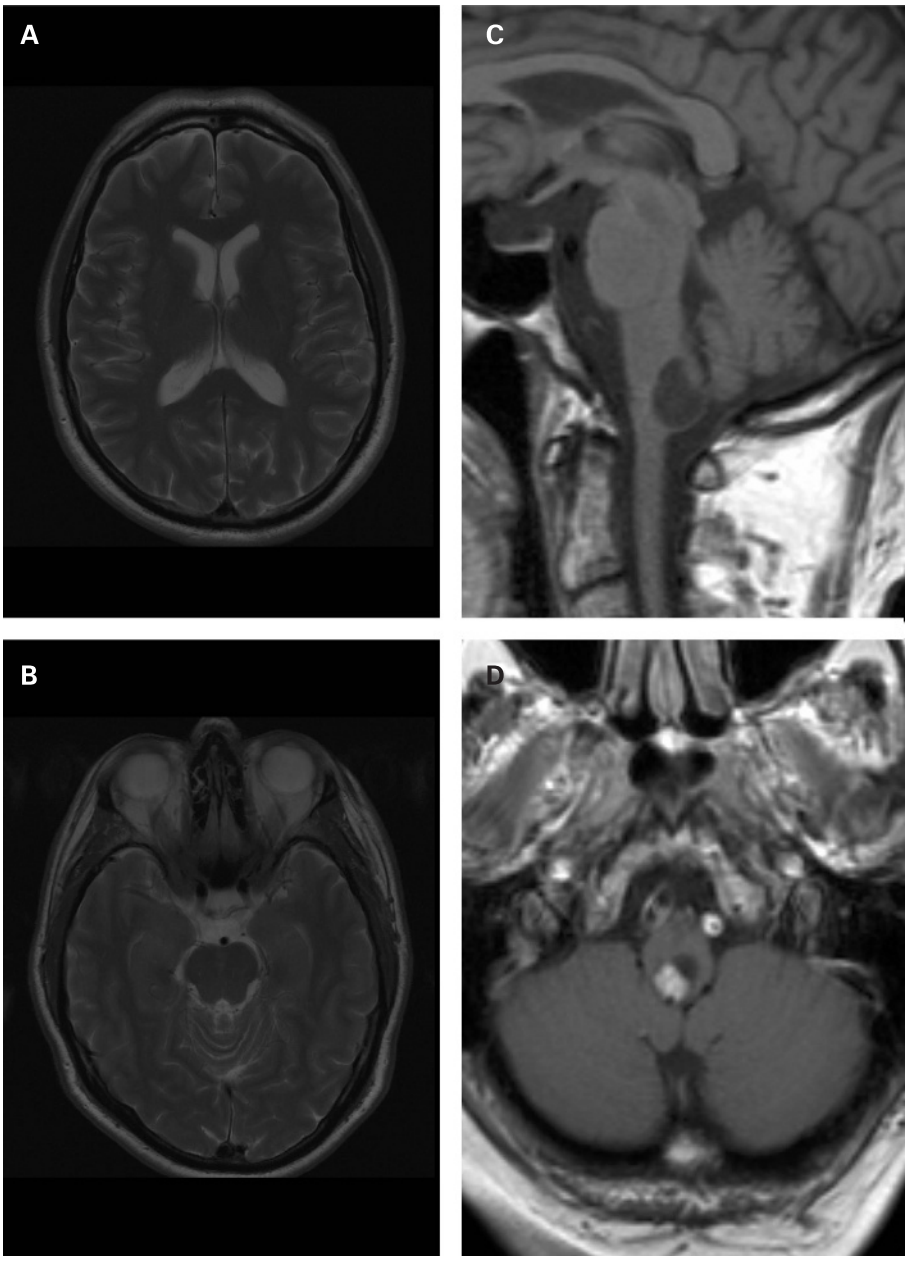

Figure 1 Magnetic resonance imaging. Axial T2-weighted fast-spin echo images (A, B) show no hydrocephalus or temporal lobe lesions. Sagittal T1-weighted spin echo (C) and post-gadolinium axial T1-weighted spin echo (D) images show a cystic lesion in the dorsal medulla oblongata with an enhancing mural nodule.

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CORRECTION

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BPC van de Warrenburg, KP Bhatia, NP Quinn. Pisa syndrome after unilateral pallidotomy in Parkinson's disease: an unrecognised, delayed adverse event? *J Neurol Neurosurg Psychiatry* 2007;**78**:329–330.

The authors of this letter would like to correct the statement that "… postoperative imaging in these three patients confirmed that the lesions were confined to the medial pallidum…" Upon further subsequent expert review of the postoperative MRI scans, it became apparent to the authors that although the medial pallidum was indeed successfully lesioned in all three patients, in each of them the lesion was in fact more extensive. In patient 1, the surgical lesion extended into the external globus pallidus and putamen; in patient 2 into the internal capsule; and in patient 3 into the external globus pallidus, with lesions further rostrally into the area of the sella media. Nevertheless, the key issue of whether the Pisa syndrome in these patients was related solely to their advanced stage Parkinson's diesease or to the surgical lesion in the medial pallidum (and beyond) remains unsettled. Therefore, the authors' invitation to report on the very long-term follow-up of pallidotomy patients in relation to the localisation of the surgical lesion still stands. The authors apologise to the readership for this unfortunate and unintended error.

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Notes