Marked reduction of tremor in essential tremor after putaminal infarct


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LETTER

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Essential tremor (ET) is the commonest movement disorder in adults. However, the precise localisation of the primary pathology is poorly understood. ET is not generally believed to be associated with significant basal ganglia dysfunction, although an early PET study demonstrated an association with overactivation of not only the cerebellum but also the contralateral striatum (as well as thalamus and sensorimotor cortex). Neurophysiological studies suggest the involvement of a central oscillator within the olivocerebellothalamicocortical circuit. In line with this, vascular lesions of the cerebellum, pons, thalamus, corona radiata and frontal cortex have been reported to result in unilateral resolution of tremor. To date, the commonest target for control of tremor using stereotactic lesions or deep brain stimulation (DBS) surgery is the thalamic ventral intermediate (Vim) nucleus (the cerebellar-receiving area of the thalamus). Our report of a case of putaminal ischaemic stroke resulting in marked improvement of contralateral ET suggests that the basal ganglia may also be important in the pathogenesis of ET.

CASE REPORT

The patient is a 75-year-old left-handed man who was first seen in our clinic in 1993, aged 60. Symptoms began at age 10 with right-hand tremor, but the left hand soon became involved. Many activities were affected, but writing was the most impaired. Subsequently, he developed head and voice tremor. Alcohol ameliorated the tremor, and his mother and two brothers had bilateral hand tremor (but there was no family history of Parkinson’s disease or other movement disorders). There was some benefit from propranolol (80 mg/day), but primidone, phenobarbital, benzodiazepines and gabapentin were either ineffective or poorly tolerated. Botulinum toxin injections were ineffective for the hand tremor, which became progressively more disabling (eg, unable to write or drink from a cup). In 2000, he underwent right Vim DBS surgery. Soon after the surgery, he presented with a marked thalamotomy-like effect and an excellent improvement in the left hand tremor. DBS was eventually switched on in 2003 because of recurrence of left hand tremor, with satisfactory tremor control. Stimulation did not provide ipsilateral benefit. Optimal stimulation parameters were 3.6 V/90 μs/185 Hz and contacts 2– and 3+. His examination (with stimulation OFF) in June 2005

Figure 1  Spiral drawing (right hand) prior to (A) and after (B) stroke. (C) Poststroke (July 2009); the worst of 10 spirals drawn in 1 day is shown. (D, E) MRI (T2-weighted and FLAIR sequences) demonstrating a small lacunar infarct (solid arrows) in the posterior part of the left putamen (dashed arrows indicate the site of deep brain stimulation electrode in the right thalamus).
is shown in video 1 (right-hand spiral drawing shown in figure 1A). He demonstrated marked postural and action tremor of the right upper limb with a Fahn–Tolosa–Martinez Tremor Rating Scale (TRS) score of 18/28 (total of scores for right upper limb rest tremor (range 0–4), postural tremor (0–4), action tremor (0–4), drawing spirals and lines (0–12), and pouring water (0–4); higher scores indicative of worse function), mild head tremor and moderate voice tremor. His TRS activities of daily living (ADL) score were 8 (range 0–27; with higher scores indicative of worse function). He considered undergoing further surgery for the right-hand tremor.

However, in December 2005, he suffered a stroke, with sudden onset of mild right hemiparesis (National Institutes of Health Stroke Scale score of 4). Right-hand tremor was also acutely improved. CT scan of the brain performed acutely did not show any evidence of haemorrhage. His weakness had almost completely resolved by 1 month poststroke. Propranolol was reduced to 10 mg/day. Video 2 and figure 1B, taken in June 2008, show only slight postural and action tremor in the right upper limb (TRS score 6/28; ADL score 3). Head and voice tremor were unchanged. No limb weakness was detectable. A 1.5 T brain MRI showed a small lacunar infarct in the posterior part of the left putamen but was otherwise unremarkable. At the last follow-up in July 2009, the TRS ADL score was again only 3. The patient continues to take propranolol 20 mg/day, but now mainly for treatment of hypertension.

Further investigations to exclude significant involvement of the corticospinal tract were undertaken. Repeat MRI using thin (1.5 mm) slices showed minor extension (spanning 0–3 mm) of the left putaminal lesion out of the postero-medio-superior aspect of the putamen into the adjacent white matter (internal capsule) (figure 1C). DBS parameters have remained the same over 6 years. The patient continues to take propranolol 20 mg/day, but now mainly for treatment of hypertension.

We can only speculate on the mechanism(s) involved in our patient. One hypothesis is that tremors may arise from an imbalance between basal ganglia and cerebellar outputs.7 According to this theory, damaged cerebellar outflow in ET may produce an imbalance that is normalised by eliminating the surviving (tremor-generating) output from the basal ganglia—as might have occurred in our patient consequent to his stroke. Although the supporting data for this hypothesis are poor,9 recent studies have revealed evidence of cerebellar degeneration in ET (with some authors postulating that the cerebellar hypermetabolism demonstrated on functional imaging2 may in fact represent a compensatory response to the neurodegenerative process);10 other authors have suggested that the antitremor effect of Vim DBS may involve disruption of pallido-ocular fibres in the subthalamic area.9 10 While the present study is only of a single patient, it raises the intriguing possibility of surgically targeting the posterior putamen to treat contralateral ET.

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Supplementary videos are published online only. To view these files please visit the journal online (http://jnnp.bmj.com).

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