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“Rubral” tremor after thalamic haemorrhage

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Abstract Tremors in association with thalamic haemorrhage have been infrequently reported, and delayed rubral tremor as a complication of such an event is quite rare. We describe a patient with a combined resting-postural-kinetic tremor due a thalamic haemorrhage. Magnetic resonance imaging showed evidence of a subthalamic involvement but failed to reveal any mesencephalic lesion. Five years after the original stroke

there was rapid and almost complete suppression of her abnormal movements, probably related to an ischaemic capsular lesion. Involuntary movements, which resemble rubral tremor, can be due to lesions upstream of the rubral and nigral out-flow system.

Key words Movement disorders
Rubral tremor · Thalamic
haemorrhage

Introduction

“Rubral” tremor is an unusual type of tremor which includes a resting, postural and kinetic component. Characteristically present at rest, it becomes more pronounced during maintenance of a fixed posture and further increases in amplitude with intentional voluntary movements.

Most patients with rubral tremor have had clinical, pathological or radiographic evidence of lesions in the upper brain stem, and causes for the sporadic cases reported in the literature include tumours [8], vascular insults [3, 5, 8], head trauma [1, 13], multiple sclerosis [4], arteriovenous malformation [6] and toxoplasma abscess [9].

Tremors in association with thalamic haemorrhage have been infrequently described [10, 14], and rubral tremor as a late complication of such an event has not been reported previously. Therefore, we report a patient with radiographic evidence of thalamic-subthalamic haemorrhage who subsequently developed a combined rest-

ing-postural-kinetic tremor in the upper extremity contralateral to the side of the lesion. Magnetic resonance imaging (MRI) failed to reveal any mesencephalic damage. The patient had a marked decrease of her abnormal movements after a later ischaemic vascular insult.

Case report

This 53-year-old, right-handed, previously hypertensive woman was in good health until April 1986 when she suffered the acute onset of right hemiparesis and was admitted to another hospital. Her speech was dysarthric and nonfluent. She remained obtunded and confused for about 3 days. Other details of the neurological examination at that time are not available. Over the following weeks speech disturbance and hemiparesis subsided, and when she was discharged from the hospital she was able to walk and talk. One month after the acute episode the patient developed involuntary movements of the right arm which worsened over the next few months until she was admitted to our institute in October 1986.

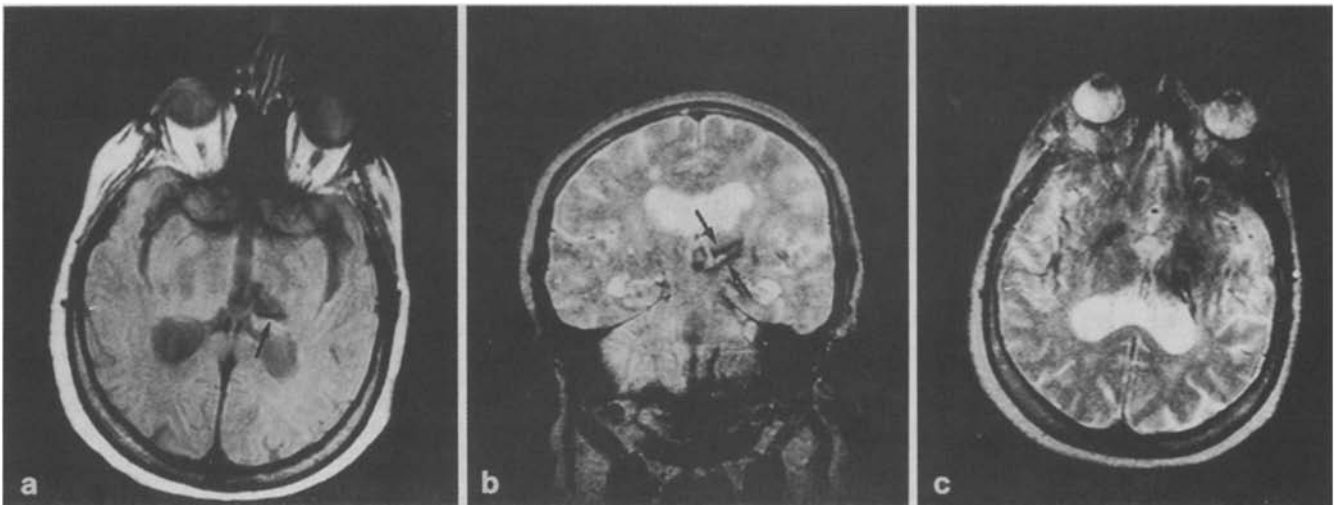
On admission she was alert, oriented, and had normal speech. Neurological examination revealed no motor weakness or sensory deficit. Gait was normal and there were no signs of cerebellar dysfunction. Deep tendon reflexes were increased on the right and the plantar response was flexor bilaterally. The pupils were slightly



Fig. 1 Handwriting of patient, attempting to sign her name

anisocoric and upgaze was limited. There was a resting tremor in the right upper extremity with a frequency of 2.5–3.0 Hz involving flexion and extension of the fingers, hand, and forearm, as well as pronation and supination of the forearm. The tremor was also present with posture and became pronounced during intentional voluntary movements. The hand tremor caused marked functional interference with the activities of daily living, especially writing (Fig. 1). A small amplitude of resting tremor was also observed in her right foot which was intermittent and not present during walking. Palatal myoclonus, myoclonic jerking and dystonic postures

Fig. 2 a–c MRI: **a** Axial section through the inferior thalamus and **b** coronal section through the posterior thalamus (TR = 1600, TE = 30, 100 ms respectively) illustrating a lesion in the left thalamus and subthalamus. The *arrows* point to an area of pronounced signal void consistent with haemosiderin deposition from an old haemorrhage. **c** A more rostral axial section T2-weighted image shows that the posterior part of the left internal capsule is unaffected



were not observed. Cranial computed tomography (CT) showed an irregular area of decreased density in the left posterior thalamus that was assumed to represent an old haemorrhagic lesion. MRI confirmed that the patient had had a thalamic haemorrhage with caudal extension into the subthalamic area (Fig. 2 a, b). There was no MRI evidence of mesencephalic damage (Fig. 3 a). Neither CT nor MRI showed any lesion of the posterior limb of the internal capsule (Fig. 2 c).

In May 1991, i.e. approximately 5 years after the onset of the abnormal movements, her family noticed the sudden disappearance of the tremor. On neurological examination, 10 days later, there was a mild right hemiparesis, more severe in the arm than in the leg, with hyperactive reflexes and extensor plantar response. Exteroceptive somatic sensation was preserved with right tactile extinction on double simultaneous stimulation. Position sense and graphaesthesia were impaired in the right hand. There was minimal tremor at rest in the right upper limb, and only right dysmetria with the finger-to-nose manoeuvre. However, a mild intentional tremor of the right hand was observed when the patient attempted to write. When she walked, she carried her right arm flexed at the elbow and showed an intermittent moderate amplitude tremor (4–5 Hz). A new MRI study disclosed, in addition to the old thalamo-subthalamic lesion, multiple patchy subcortical areas of increased signal intensity localized in the periventricular white matter (present in part at the first MRI study), left cerebellar hemisphere, and left internal capsule (Fig. 4). Again MRI confirmed that the red nucleus and the areas around it were not involved (Fig. 3 b).

Discussion

The patient exhibited the late appearance of unilateral tremulous movements as the major manifestation of contralateral posterior thalamic haemorrhage confirmed by MRI. There was good recovery of speech and motor function, and when she was admitted to our hospital no sensory impairment or cerebellar dysfunction were observed.

Our patient's tremor had many features of what has been described in the literature as midbrain or rubral tremor [7]. There is no complete agreement regarding the definition of rubral tremor – whether this disorder is a

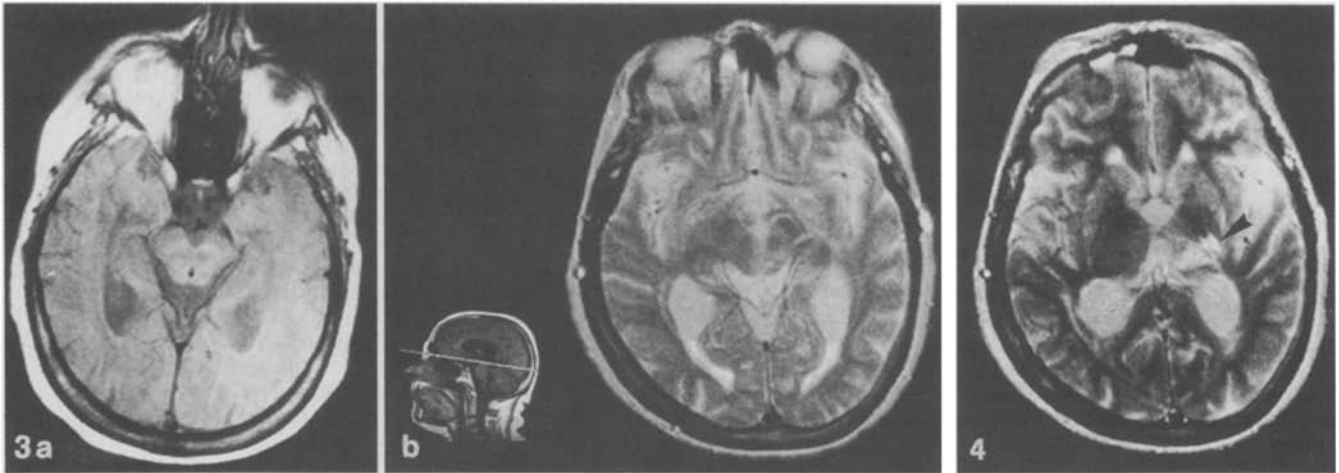


Fig. 3 a, b Transaxial MR images through the upper mesencephalon (TR 1600, TE 30 ms; TR 2000, TE 110 ms respectively) showing that the area around the red nucleus is intact

Fig. 4 T2-weighted axial MR image (TR/TE = 2000/110 ms) demonstrates areas of high signal intensity in the left thalamus and left posterior limb of the internal capsule. The more intense high signal abnormality from the capsular area (*arrowhead*) is consistent with recent infarction

single tremor or a combination of two distinct tremors. Some authors believe that a rubral tremor is actually a cerebellar tremor associated with a parkinsonian tremor. The kinetic tremor component is thought to be related to lesions involving any part of the dentato-rubro-thalamic pathway, whereas the rest component requires involvement of the ascending nigrostriatal pathway [5]. Midbrain tremor is commonly referred to as rubral tremor because of the presumed involvement of the red nucleus. However, lesions of this nucleus are not essential for the development of the tremor [11], whereas lesions in the environs of the red nucleus may lead to a rubral tremor whether or not the nucleus is damaged. It appears that the tegmental area around the nucleus is particularly important in the genesis of the tremor [13].

The radiographic data from our patient failed to reveal any mesencephalic lesion. CT and MRI showed evidence of an old haemorrhagic lesion restricted to the posterior thalamus and rostral subthalamus. Therefore, the present case shows that rubral tremor is not exclusively due to selective damage of structures located in the upper brain stem, and that a well-placed lesion upstream of the rubral

and nigral outflow can cause a tremor of similar type. In spite of the lack of pathological confirmation, we believe that the involvement of the subthalamic area was critical in the genesis of the tremor, because probably at this level both projection pathways from the dentate to the ventrolateral thalamic nucleus and the dopaminergic nigrostriatal pathway were interrupted.

Tremor is abolished by surgical intervention on the motor cortex [2], by pyramidotomy at the lower brain stem [15] or after cerebral infarct producing a hemiplegia [12]. These observations indicate that tremor is driven by discharge in the motor cortex transmitted via neuronal pathways traversing the internal capsule.

Five years after the original stroke, this patient suddenly became hemiparetic again and had almost complete suppression of her abnormal movements. A new MRI study revealed multiple patchy white-matter foci of increased signal intensity; most of these had been seen in the first MRI. The small bilateral lesions involving the white matter adjacent to the lateral ventricles could be regarded as incidental subcortical lesions associated with age and hypertension, and cannot account for the events described. Conversely, the hyperintense area identified in the posterior limb of the internal capsule probably represents an ischaemic lesion and may explain the recent sensorimotor deficit, as well the disappearance of the tremor, since the involvement of the pyramidal tract presumably caused interruption of the tremor impulses in their downward transmission.

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