Spasmodic torticollis relieved by removal of a cervical foreign body

Sir: The neurological basis of spasmodic torticollis is obscure and consistently effective treatments have proved elusive. A case is presented of a man whose spasmodic torticollis appeared to be attributable to a foreign body embedded in the neck and was relieved by its removal.

A 32-year-old man had been stabbed in the left side of the neck with a glass 14 years previously. During the subsequent seven years he received further stab wounds to the left chest and right hand and a nasal fracture. Septorhinoplasty was performed when he was aged 28 years. At 29 years the site of the injury to the neck became uncomfortable and he found he could relieve the "annoying sensation" by turning his head to the left. An intermittent stabbing pain later added to the continuous ache and the torticollis became concomitantly more severe to the extent that it interfered with his work as a salesman. The movements had become involuntary and he described them as starting with a twitch. The torticollis remained truly spasmodic and he found it increasingly difficult to override the movement voluntarily; at times his neck would turn to, and lock in, the fully rotated position with his face to the left. The patient was convinced that something was embedded in his neck, but was referred for psychiatric treatment, acupuncture, osteopathy, local injections of lignocaine and physiotherapy including traction, all without benefit, prior to neurosurgical referral. He was on no medication.

Examination showed him to be a generally fit man with a traumatic scar in the posterior triangle on the left side of the neck overlying a firm palpable nodule. The right sternocleidomastoid muscle was hypertrophied relative to the left (fig) and photographs showed that this asymmetry had not been present seven years earlier. There was a persistent tendency for his face to turn to the left, often abruptly, despite a constant conscious effort by the patient to maintain a neutral position. The frequency and amplitude of the movements were exacerbated by both motor and psychological loading. There was no retrocollis or lateral neck flexion, titubation or trunk torsion, no facial dyskinesia and no disturbance of ocular movements, pupillary responses, speech or swallowing. None of the limbs was dystonic and there was no tremor. His gait was normal and there were no cerebellar or long tract signs. Radiographs showed a glass fragment beneath the scar and this was removed under general anaesthesia from its position 2 cm below the surface. By five weeks after operation the torticollis was apparent only when walking, could easily be overridden and his neck no longer "locked".

By thirteen months it had almost completely resolved subjectively with only a feeling of tightness in the shoulders and base of the neck while driving. Objectively, his neck movements were full and free in all directions, but very slight turning movements were still apparent under stress. The muscular asymmetry was less marked than preoperatively.

Torticollis may be familial1 – 3 and is considered by some to be a variant of dystonia muscularum deformans.4 4 It may be associated with writer's cramp5 and may occur after encephalitis,6 7 after neuroleptic drug administration8 and in cases of asymmetrical Parkinson's disease after dopamine agonist administration.9 10 Cases have been attributed to posterior fossa tumours,11 syringomyelia with cord tumours,12 and thyrotoxicosis13 and it has long been recognised that torticollis may follow a dynamic injury to the neck.14 But the aetiology is often obscure.6 15 Although torticollis can be reproduced experimentally in animals by lesions in the midbrain tegmentum,16 –18 and there is some experimental evidence implicating altered nigro-striatal dopaminergic transmission,19 20 the pathophysiology of torticollis in man remains unknown.7 It might be proposed that the torticollis in the present case was not directly related to the foreign body but was exacerbated by the stress of his being regarded as "functional". The lack of improvement whilst waiting for surgery and the extent of the relief following surgery refute this. An explanation in terms of direct local irritation by the foreign body is improbable as the excess activity and subsequent muscular hypertrophy were contralateral. Even if he should later develop the full syndrome of dystonia muscularum deformans, he was dramatically relieved of his torticollis for at least thirteen months following surgery. Whilst spontaneous remissions are recognised in dystonia, particularly in the adult-onset type,14 an alternative hypothesis is that the torticollis was induced and maintained by afferent input from the presence of the foreign body. I am grateful to Professor ES Watkins for allowing me to report this case.

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Fig Isometric contraction revealing relative hypertrophy of the right sternocleidomastoid muscle.

References
Letters


Accepted 3 February 1986

Immuno-gold silver staining in the diagnosis of herpes encephalitis

Sir: The practical difficulties of obtaining a definitive diagnosis of herpes encephalitis are well documented,1 and the diagnostic method based on the detection of a central nervous system immune response to Herpes virus simplex (HSV), previously reported from this laboratory,2 is not wholly reliable in the early stages of the disease (that is, less than 10 days after onset). Until more reliable non-invasive diagnostic tests become available, brain biopsy (albeit an increasingly unpopular procedure3) will remain the most reliable means of achieving an early diagnosis.

Brain material may be examined in a variety of ways4 including electron microscopy, immunofluorescence, immunoperoxidase and peroxidase anti-peroxidase staining. We have recently applied a new histochemical method, immuno-gold silver staining (IGSS)5 to the detection of virus antigen in such specimens. In this procedure the specimen is overlaid with a rabbit anti-HSV serum (antiserum prepared in this laboratory to HSV strain Syn 17 +), followed by anti-rabbit colloidal gold conjugate (Janssen Life Sciences Products, Belgium). The presence of virus antigen is then revealed as distinct black staining deposits by silver development.

The procedure although slightly more time consuming (about 3½ hours) than staining by immunofluorescence or immunoperoxidase is straightforward and appears to offer considerable advantages in terms of ease of reading and sensitivity of detection.

In the evaluation of brain material examined post-mortem and for studies related to pathogenesis, this procedure provides advantages over methods used to date. The sensitivity of the method will allow improved investigation of viral spread within the brain. In addition, in comparison with other histochemical methods, the IGSS method is able6 to detect antigen in tissue where there has been a significant reduction of antigenicity during fixation or paraffin processing.

The figure shows an area of virus infection within the temporal lobe, of a patient with herpes encephalitis, stained using the IGSS method. Controls in which specimens were “stained” with the primary antiserum replaced by normal rabbit serum showed no reactivity. Also there was no background interference which might have hindered interpretation. Specific staining, predominantly of the nuclear and cytoplasmic membranes of both neurons and glial cells, is observed with IGSS. The distribution of “stain” correlates well with areas of viral activity detected by peroxidase-anti-peroxidase or immunofluorescence.

Current use in this laboratory leads us to believe that IGSS will be the method of choice as long as biopsy material is to be used in the diagnosis of herpes encephalitis. The full potential of this method will be realised when IGSS is more widely applied.

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IGSS staining of a 5 μm section of paraffin embedded brain (× 350). The patient died from herpes encephalitis, brain biopsy proven (culture HSV type 1) and immunofluorescence positive). Grey matter of left temporal cortex with particulate staining of four neurons, this is present in the cytoplasm but in the neuron in the lower right (arrow head) there is also nuclear staining. Neuron in upper field (arrowed) not stained.
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*J Neurol Neurosurg Psychiatry* 1986 49: 1208-1209
doi: 10.1136/jnnp.49.10.1208

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