Low prevalence of HTLV-1 antibodies in the serum of patients with tropical spastic paraplegia from the Ivory Coast

A high prevalence of human T-lymphotropic virus type I (HTLV-1) antibodies has been reported in the serum of patients with tropical spastic paraplegia (TSP), from various parts of the world. This disease is also present in West Africa where, as in other tropical regions, no obvious aetiology was discovered.

To discover the possible link between TSP and HTLV-1 antibodies in West Africa, the serum of 20 patients from the Ivory Coast was collected. All patients fulfilled the clinical diagnostic criteria previously described for TSP. Other causes of paraparesis were excluded by clinical and paraclinical inves-

tigations. The presence of HTLV-1, HIV-1 and HIV-II antibodies was determined by ELISA and western-blot methods. The results are summarised in the table. One patient was positive only for HTLV-1, two patients were positive for HTLV-1 and HIV-II, one was positive for HIV-II and one was positive for HIV-I and HIV-II. High 1HTLV-1 antibody titres were found ranging from 1/5000 to 1/10 000.

The total prevalence of HTLV-1 positivity among TSP patients was 15%. This observation contrasts with the low prevalence of HTLV-1 positivity reported in the other tropical regions, particularly in the Seychelles (85%), and in Martinique (59%).

The seropositivity of HTLV-1 among healthy controls in the Ivory Coast (1.6%) is similar to that observed in the French West Indies (2%).

These findings confirm that in West Africa antibodies against HTLV-1 antibodies other than HTLV-1 are present in the serum of patients with TSP, as we reported previously. The lack of HTLV-1, HIV-I and HIV-II antibodies in 75% of patients with TSP indicates that different T-cell subpopulations may be linked to TSP in West Africa, such as other viruses, toxins or malnutrition.

Our results underline the need for a larger study of TSP in Africa to evaluate the role of various aetiological factors with the geographical distribution of the disease throughout the continent.

This work was supported by a grant from The French Ministry of Cooperation (DPR RS423).

Table: Results of HTLV-I, HIV-I and HIV-II antibodies in the serum of 20 TSP patients from the Ivory Coast (ELISA and western-blot methods).

| Patient | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | 17 | 18 | 19 | 20 |
| Titre  | + | + | + | + | + | + | + | + | + | + | + | + | + | + | + | + | + | + | + |

Wegener’s granulomatosis presenting as peripheral neuropathy: diagnosis confirmed by serum anti-neutrophil antibodies

The case described by Kirker, Keane and Hutchinson illustrates that neuropathy may occur early in Wegener’s granulomatosis with the absence of more classical pulmonary and renal findings, leading to delay in diagnosis. Even if the diagnosis is suspected, histopathological confirmation may be difficult. We describe a patient presenting with rapidly progressive peripheral neuropathy and inconclusive biopsy findings for whom the demonstration of serum autoantibodies to neutrophil cytoplasmic antigen (ANCA) permitted early diagnosis and therapy.

A 57 year old man developed rapidly progressive numbness and weakness of both hands and the right leg over a period of 10 weeks. He had right wrist and foot drop and marked wasting of both hands. Right elbow, wrist, hand, ankle and foot movements were MRC grade 2–3, limb power elsewhere was grade 5. Pin sensation and proprioception were diminished over both hands, right foot and the lateral aspect of the right leg; other sensation was unimpaired. He had a vesiculopapular rash on his elbows and hands but no other abnormality, and urinalysis was normal.

Urea, electrolytes, haemoglobin and chest radiograph were normal. White cell count was 19.6 x 10^9/L, 74% neutrophils, ESR 38 mm/hour, and serum C-reactive protein 45 mg/l. Serum alkaline phosphatase was 1031 U/l, alanine transaminase 211 U/l, and aspartate transaminase 73 U/l. Urinary creatinine clearance was moderately reduced at 81 ml/min. Rose-Waaler test was positive (titre 1: 1280). Serum B12, blood lead, abdominal ultrasound scan and CSF were normal. Screening for porphyria, hepatitis viruses and antinuclear and antimitochondrial antibodies were negative, digital renal arteriography showed no aneurysms, and the only abnormality on CT scanning of the skull, thorax and abdomen was fluid in both maxillary sinuses. ENA assessment revealed a granular mass on the right postnasal space. Biopsies of this area and of the skin lesions showed non-specific chronic inflammation. Despite unhelpful histopathological findings, Wegener’s granulomatosis was confirmed by the presence of serum ANCA was confirmed on indirect immunofluorescence (titre 1: 80).

The patient was started on cyclophosphamide 2 mg/kg and a reducing dose of prednisolone. Six months later right wrist extension and finger movements were grade 3; limb power was otherwise normal. Sensory impairment was limited to the fingertips and big toe. ESR, C-reactive protein, white cell count and creatinine clearance were normal, Rose-Waaler negative, and ANCA not detectable.

Histopathological confirmation of Wegener’s granulomatosis may be difficult. Parlevliet et al. obtained a histological diagnosis in only 2 of 11 patients with typical symptoms and signs of the disease. While a presumptive diagnosis is acceptable when the presentation is typical, the decision to start cytotoxic therapy with its attendant morbidity is difficult in atypical cases.

Savage et al. showed that ANCA was not present in a small series of patients with polyarteritis nodosa and Churg-Strauss syndrome, but was highly sensitive and specific for Wegener’s granulomatosis and micros-
copic polyarteritis. Both conditions require prompt treatment with cyclophosphamide, and the distinction between the two may be semantic only. We believe that early treatment with cyclophosphamide in this case, based on the finding of serum ANCA, prevented further deterioration in renal function and possible pulmonary involvement.

Serum ANCA is a useful marker for patients with unusual neuropsychiatric presentations, particularly when the poor faces and picture memory scores of their patient were simply secondary to a visuoconstructual impairment, as suggested by the patient's low Performance IQ, rather than related to a global memory disorder per se.

There is one important feature of AK patients' memory functioning which was not covered in the report by Feinstein and Ron, namely retrograde memory performance. In the case of their patient, this is critical for two reasons. Firstly, significant retrograde memory loss appears to be a hallmark of the AK amnestic syndrome. In my review of memory disorders, I was unable to find a single case of an AK patient with marked anterograde memory impairment who did not also have poor retrograde memory functioning. Although retrograde memory deficits may present in some patients with chronic-progressive multiple sclerosis, the pattern of memory loss differs in the two conditions—a temporal gradient in the memory loss is present in AK patients, whereas most test conditions, and severe retrograde amnesia in the absence of generalised cognitive dysfunction is a feature of the global memory disorder of many AK patients, but less so in the case of patients with multiple sclerosis, where remote memory loss in patients without significant dementia is only mild or moderate in severity.

Secondly, marked retrograde memory loss is a characteristic which also distinguishes AK patients from those with neuropsychological deficits resulting simply from chronic alcohol consumption alone. Feinstein and Ron's brief clinical observations of their patient's retrograde memory functioning suggest a rather patchy retrograde amnesia, with the patient reported as having poor memory for past personal events but intact memory of more recent events. This is consistent with previous medical history, and if this suggestion of a limited retrograde amnesia was upheld by formal assessment it might be more in keeping with multiple sclerosis rather than an AK syndrome. Since marked anterograde memory deficits and other cognitive dysfunctions can occasionally be present in patients with multiple sclerosis who do not have severe neurological disability, this diagnosis alone could explain most of the evidence reported by Feinstein and Ron.

While it is quite possible that a proportion of the anterograde memory impairment shown by Feinstein and Ron's patient may have been due to chronic alcohol abuse, it would have been useful (allowing for the normal gamma GT) to know the duration of abstinence to exclude the role of any recent, acute effects of alcohol consumption on memory functioning. It may also have been useful to document brain stem auditory evoked responses to examine the possibility of differences which could have existed between some Wernicke-Korsakoff patients and those with multiple sclerosis.

In the case of other neuropsychological features of the case presented by Feinstein and Ron, I would agree that the absence of IQ differences, and the evidence of nominal dys-
Wegener's granulomatosis presenting as peripheral neuropathy: diagnosis confirmed by serum anti-neutrophil antibodies.

W Dickey and W J Andrews

*J Neurol Neurosurg Psychiatry* 1990 53: 269-270
doi: 10.1136/jnnp.53.3.269-a