zones. We concluded that the choice of structures for exploration had been in error. Interictal positron computed tomography showed a zone of relative hypometabolism in the right frontal lobe but the degree of depression was not statistically significant.

Three months later a second stereo-EEG exploration was performed. Having eliminated a location for the epileptic focus in the temporal lobe, the mesial frontal region and the cingulate gyrus, we explored the frontal orbital region and the frontal pole. Five electrodes were inserted through the frontal lobes (fig). Frequent interictal spikes and spike-waves were recorded consistently in the frontal orbital region without extension to any ipsilateral cortical zone, but with occasional propagation to the contralateral orbital frontal region. Many paroxysmal discharges unaccompanied by clinical manifestations were found to occur in a very restricted zone during sleep. Electroclinical seizures were always characterised by repetitive spike discharges located only in the right orbital frontal region. Elsewhere flattening was observed as previously (fig). Because irritative and epileptic foci were restricted to the same orbital frontal region, we performed a cortical resection of the orbital gyri, gyrus rectus and area subcallosa. The patient has now been seizure free for 18 months since the operation. Histological examination of the excised brain showed no abnormalities.

Complex partial seizures may originate in the frontal lobe^{3,4} and to distinguish their origin from those of temporal lobe origin the patterns of the discharge spread seem to be more useful than the characteristics of the automatisms themselves.^{4,5} Geier *et al*³ have pointed out the high frequency of motor manifestations: deviations of the head and eyes (86, 4%), clonic and or tonic manifestations (77, 3%), and phonatory manifestations (86%). Topographic localisation of frontal lobe epilepsy by routine EEG is often difficult.⁶ For these reasons nasoethmoidal and supra-orbital electrodes, electrocorticography and sometimes stereo-EEG have been used.^{6,7}

Usually, when an orbital frontal epileptic focus has been suspected, a complete frontal lobectomy has been performed but few cases with follow up have been reported. Niedermeyer *et al*⁸ reported one case whose seizures improved and Tharp² one whose seizures stopped. Rasmussen⁹ studied 40 cases with frontal lobe epilepsy who remained seizure-free for a minimum of five years after surgery. The clinical patterns were "absence" (35%) and simple or complex automatisms (30%). Neither the origin of the discharges nor the correlations

between the extent of resection and the type of seizure were specified. Ludwig *et al*¹ presented four patients with seizures or probably orbital frontal origin: one patient had been operated by orbital frontal resection which was followed by relative improvement. A temporal and orbital frontal resection was performed in a second case but follow up was not reported. Our case seems to be the only one of orbital frontal epilepsy in which a resection restricted to the orbital gyrus has been reported and followed by cessation of seizures. Similar localisation of inter-ictal spikes and ictal discharge combined with the lack of spreading to other structures led us to choose a restricted resection.

We conclude that no clear-cut clinical pattern is diagnostic of a frontal origin of complex partial seizures. Moreover, the discharges are rarely limited to the orbital frontal zone; more often they spread to other frontal or temporal regions. When such an origin is suspected stereo-EEG can be of real diagnostic value in delimiting the epileptic focus.⁷

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References

- Ludwig B, Ajmone Marsan C, Van Buren J. Cerebral seizures of probable orbito-frontal origin. *Epilepsia* 1975;16:141-58.
- Tharp BR. Orbito-frontal seizure. A unique electroencephalographic and clinical syndrome. *Epilepsia* 1972;13:627-42.
- Geier S, Bancaud J, Talairach J, Bonis A, Szikla G, Engelvin M. The seizures of frontal lobe epilepsy. A study of clinical manifestations. *Neurology* 1977;27:951-8.
- Williamson PD, Spencer DD, Spencer SS, Novelly RA, Mattson RH. Complex partial seizures of frontal lobe origin. *Ann Neurol* 1985;18:497-504.
- Brey R, Laxer KD. Type 1/2 complex partial seizures: no correlation with surgical outcome. *Epilepsia* 1985;26:657-60.
- Quesney LF. Seizures of frontal lobe origin. In: Pedley TA, Meldrum BS, eds. *Recent advances in Epileptology*. London: Churchill Livingstone, 1986:81-110.
- Bancaud J, Talairach J, Bonis A, *et al*. La stéréo-électroencéphalographie dans l'épilepsie. Paris: Masson 1985:321.
- Niedermeyer E, Walker AE, Burton C. The slow spike-wave complex as a correlate of frontal and fronto-temporal post-traumatic epilepsy. *Eur Neurol* 1970;3:330-46.
- Rasmussen T. Characteristics of a pure culture of frontal lobe epilepsy. *Epilepsia* 1983;24:482-93.

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Intermittent pyramidal claudication as presenting and sole symptom in multiple sclerosis

Sir: Upper-motor neuron gait disturbance, only appearing on strenuous exercise and relieved by rest, is a rare variant of neurogenic intermittent claudication and has been known as "intermittent claudication of the spinal cord". More appropriately, however, the syndrome may be called Intermittent Pyramidal Claudication (IPC). It manifests itself as a feeling of discomfort, indolent heaviness and weakness in the legs with a spastic gait-pattern, after a period of walking. Upper-motor neuron signs and symptoms may appear on exertion and disappear on rest. During rest or mild exercise, patients are typically free of complaints and symptoms. IPC is generally thought to be due to transient ischaemia of the spinal cord, secondary, for example, to arteriosclerosis, (syphilitic) arteritis, spinal cord compression or vascular malformations.¹⁻³ Multiple sclerosis is usually not mentioned as a cause. However, in the pertinent literature on multiple sclerosis, IPC is mentioned as "a common presenting symptom, which usually quickly progresses into persistent paresis".⁴⁻⁶ Remarkably, informative case reports or reviews on this subject are scarce. This prompted us to present the case histories of two patients with IPC which proved to be the initial and sole symptom of multiple sclerosis for 6 and 4½ years.

Patient 1: Five years before admission, this 52 year old construction worker noticed weakness, clumsiness and a heavy feeling in both legs, predominantly on the right, after walking or bicycling for about 45 minutes. Then he was likely to stumble and fall. The symptoms always disappeared after sitting or standing still for about 15 minutes. During rest or mild exercise he was asymptomatic. Time delay to onset of the symptoms gradually decreased. One year later, he noticed numbness and clumsiness of his right arm, on using it extensively for over an hour. These symptoms disappeared after a 30 minute rest. Two years after onset, the intermittent gait disturbance forced him to stop working. One year later, neurological assessment, however, without exertion-provocation, was normal. Five years after onset of the symptoms, he was referred to Leiden University Hospital. At that time, symptoms began after walking for about 15 minutes. Physical examination at rest revealed no abnormalities, except for absent abdominal reflexes. However, after walking for 30 minutes his gait became spastic and awkward. Neurological examination then revealed a brisk biceps jerk, inex-

haustable ankle clonus and extensor plantar reflex, all on the right side. After a 15 minutes rest, upper-motor neuron signs and symptoms had disappeared. Routine laboratory tests including syphilis serology and complement studies were normal or negative. ANA and rheumatoid factor were negative. The CSF showed high IgG and one extra oligoclonal band at iso-electric focusing. Myelography, spinal angiography and cranial CT scan were normal. MRI showed multiple hemispherical white matter lesions, suggesting multifocal demyelination. Visual-evoked-responses (VER) and somatosensory-evoked-responses (SSER) showed increased latencies.

Patient 2: Six years before admission, this 58 year old book-keeper repeatedly noticed brief jerks of his right leg after walking for 10 minutes, which disappeared after resting for 5 minutes. One and a half years later he was troubled by a heavy feeling in the right arm with numbness of the first three fingers, on using it extensively. These symptoms disappeared after 30 minutes rest. Four years after onset he noticed weakness of the legs on walking for 5 minutes, which disappeared during rest. These symptoms forced him to work for half days only. Six months later, walking felt stiff and awkward with pain and tingling of the legs and an imperative micturition developed. When lying in bed he noticed jerking movements of the legs. Six years after onset of the symptoms he was admitted to our hospital. Neurological examination revealed no abnormalities except for absent abdominal reflexes. His gait was normal initially, but became spastic after walking for 1 hour and a brisk left biceps jerk, exaggerated ankle jerks and bilateral extensor plantar reflexes appeared. After a 10 minute rest upper-motor neuron signs and symptoms disappeared. Routine laboratory tests were normal. CSF examination revealed high IgG and multiple extra oligoclonal bands at iso-electric focusing. Myelography was normal. MRI revealed multiple paraventricular white matter lesions consistent with multifocal demyelination. VER showed prolongation of P100 on stimulation of the right eye, SSER showed absent cortical responses on bilateral tibial nerve stimulation and electro-oculography showed square wave jerks on eye closure.

The main features of both case histories are intermittent, exertion-induced, upper-motor neuron symptoms and signs, disappearing after rest. During rest or mild exercise both patients were subjectively and objectively asymptomatic. Normal "vasculitis tests", angiography, myelography and MRI of the spinal cord made vascular

lesions or compression of the spinal cord most unlikely. Based on the results of CSF studies, multimodality evoked potentials tests and MRI, in both cases a diagnosis of "laboratory-supported-definite multiple sclerosis" was made, according to the criteria of Poser *et al.*⁷ In the literature on neurogenic intermittent claudication, multiple sclerosis is not generally cited as a cause for IPC.^{1,2} On the other hand, textbooks on multiple sclerosis mention IPC as "a frequent early symptom of multiple sclerosis", however, without referring to well documented observations.^{4,5} Descriptions of signs and symptoms are variable and sometimes vague.^{4,5,8,9} The period during which upper-motor neuron signs and symptoms really are intermittent (that is, they completely disappear after rest) appears to be short.^{4,9} Only three short case histories could be found in the literature.^{6,10} McAlpine⁶ described two patients who, as early feature of multiple sclerosis, showed IPC for several weeks. In both cases this disappeared for about a year and then recurred as a prelude to a progressive course of the disease. Godlewski¹⁰ mentioned a multiple sclerosis patient who had IPC for 4 months, before permanent spastic paraplegia developed. Multiple sclerosis presenting with slowly progressive IPC as isolated symptom for nearly 6 years has, to the best of our knowledge, not been described before. The pathophysiological mechanism of IPC in multiple sclerosis is believed to be related to diminished motor conduction in demyelinated axons, resulting from even a slight rise of body-temperature caused by exercise.^{4,5,11} A temperature increase of only 0.5°C appears to be sufficient to produce reversible conduction-block in demyelinated fibres.¹² Indeed in our first patient, walking for 30 minutes increased tympanic temperature by 0.5°C. Obviously, comparison of pre- and post-exertion corticospinal tract conduction time¹³ in patients with IPC, may give better insight in the pathophysiological mechanism.

IPC is generally attributed to transient spinal ischaemia.¹ However, actual evidence for such causal relationship is sometimes poor.^{3,14} Conduction or angiographic evidence are rarely found. In retrospect, some of these cases might in fact have been caused by multiple sclerosis.

In conclusion, these two case histories illustrate that IPC may be a solitary manifestation of multiple sclerosis for protracted periods. A simple exertion-provocation-test may be essential to reveal the neurological substrate of the complaints and to establish the diagnosis.

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References

- Verbiest H. Neurogenic intermittent claudication. In: Vinken PJ, Bruyn GW. *Handbook of Clinical Neurology*. Vol 20. Amsterdam: North-Holland Publishing Company, 1978: 611-724.
- Jellinger K, Neumayer E. Claudication of the spinal cord and cauda equina. In: Vinken PJ, Bruyn GW. *Handbook of Clinical Neurology*. Vol 12. Amsterdam: North-Holland Publishing Company, 1972:507-47.
- Dejerine J. Sur la claudication intermittente de la moelle épinière. *Rev Neurol (Paris)* 1906;14:341-50.
- Matthews WB, Acheson ED, Batchelor JP, Weller RO. *McAlpine's Multiple Sclerosis*. Edinburgh: Churchill Livingstone, 1985.
- Kelly R. Clinical aspects of multiple sclerosis. In: Vinken PJ, Bruyn GW. *Handbook of Clinical Neurology*. Vol 47. Amsterdam: Elsevier Science Publishers 1985:49-78.
- McAlpine D, Compston ND, Lumsden JH. *Multiple Sclerosis*. Edinburgh: Livingstone, 1955.
- Poser CM, Paty DW, Scheinberg, *et al.* New diagnostic criteria for Multiple Sclerosis: Guidelines for research protocols. *Neurology* 1983;13:227-31.
- Claude H, Egger W. Quelques symptômes nouveaux de la sclérose en plaques. *Rev Neurol (Paris)* 1906;14:275-7.
- Guillain G. La sclérose en plaques. *Rev Neurol (Paris)* 1924;24:648-83.
- Godlewski S. Les claudications intermittentes médullaires et radiculaires. *L'Hôpital (Hors-série)* 1969:529-48.
- Davis FA. Axonal conduction studies based on some considerations of temperature effects in multiple sclerosis. *Electroencephalogr Clin Neurophysiol* 1970;28:281-6.
- Rasminsky M. The effects of temperature on conduction in demyelinated single nerve fibres. *Arch Neurol* 1973;28:287-92.
- Mills KR, Murray NMF. Corticospinal tract conduction time in multiple sclerosis. *Ann Neurol* 1985;18:601-5.
- Dupré ME. Claudication intermittente par artériosclérose de la moelle. *Rev Neurol (Paris)* 1914;28:498-500.

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Unexplained chronic subarachnoid bleeding and a slowly progressive neurological syndrome

Sir: Superficial haemosiderosis of the central nervous system (CNS) (subpial haemorrhage)