Letters

Orbital frontal epilepsy: a case report

Sir: Although an orbital frontal origin for complex partial seizures has been sometimes suggested, few cases have been completely documented.1,2 We report a patient in whom electroclinical correlations were obtained by electroencephalography (EEG) and stereo-EEG recordings. The disappearance of seizures after a surgical resection limited to the orbital frontal cortex confirmed the localisation of the epileptic focus.

A 29 year old male had begun to experience seizures when 10 years old. The aetiology was unknown. Neurological examination was normal. The seizure patterns did not change during the next 19 years. They were characterised by staring, by sudden and incomplete loss of contact followed by semi-purposeful automatisms, by thrashing movements if he was held, by the shouting of incoherent words and sometimes by laughter. Deviation of the head and eyes to either side seemed to mimic natural movements and were accompanied by an expression of bewilderment. The seizures were brief (lasting about 30 seconds) and ended suddenly. Most remained partial. Some secondary generalised seizures occurred during sleep, usually when the patient had forgotten his medication. After the seizures, the patient claimed awareness but, in fact, he had an amnesia for the events around the attacks. They occurred in groups of 3 to 10 per day. Sometimes he remained seizure-free for 1 or 2 weeks. All therapeutic trials were unsuccessful and this refractory epilepsy prevented the patient from finding employment in spite of a professional education.

All the routine EEG recordings showed a normal background cerebral activity. Theta rhythms and sharp-waves were noted over the right fronto-temporal region (F8–T4) and they increased during sleep. The only alteration marking the beginning of a seizure was a sudden attenuation of background activity quickly followed by movement artefacts rendering the recording uninterpretable. Computed tomography, bilateral carotid angiography and ventriculography were normal.

The hypothesis of a right fronto-temporal epileptic focus guided the choice of structures for stereo-EEG. Two electrodes (10 plots) were implanted in the right temporal lobe to explore the middle gyrus, amygdala and hippocampus. Two electrodes were located in the right frontal lobe to investigate the mesial frontal zone and the anterior part of the cingulate gyrus. Infrequent spikes occurred in the amygdala and sometimes in the mesial frontal cortex. Three spontaneous seizures were recorded, each had the same pattern: diffuse flattening and no paroxysmal discharge in the explored sites in the frontal or temporal...
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 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. Geier et al 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. For these reasons nasoethmoidal and supra-orbital electrodes, electrocorticography and sometimes stereo-EEG have been used.

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 reported one case whose seizures improved and Tharp one whose seizures stopped. Rasmussen 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 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 performed 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.

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References

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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 symptomatology 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) arteries, 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 41 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, inexc-
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