Epileptic Nocturnal Wanderings with a Temporal Lobe Origin: A Stereo-Electroencephalographic Study

Lino Nobili, MD; Stefano Francione, PhD; Francesco Cardinale, MD; and Giorgio Lo Russo, MD

Summary: To show the results of the exploration conducted with intracerebral electrodes in a patient affected by epileptic nocturnal wanderings (ENWs).

Method: The patient was investigated with long-term video-stero-electroencephalographic (SEEG) monitoring by means of stereotactically introduced intracerebral electrodes.

Results: We recorded four nocturnal seizures with typical features of ENWs. The SEE recordings demonstrated a well-localized ictal discharge always confined to the right temporal structures with secondary spread to the cingulate regions.

Discussion: Together with paroxysmal arousals and nocturnal paroxysmal dystonia, ENWs has been considered as a manifestation of the nocturnal frontal lobe epilepsy. Our investigation and the result of surgical outcome in this patient indicate that in some cases such episodes could have a temporal-lobe origin.

Key words: Epileptic nocturnal wanderings; frontal lobe epilepsy; temporal lobe epilepsy; intracerebral electrodes; somnambulism; sleepwalking

INTRODUCTION

PAROXYSMAL MOTOR BEHAVIORS OCCURRING DURING SLEEP ARE CLASSIFIED EITHER AS SLEEP-RELATED Epileptic seizures OR AS PARASOMNIAS. In 1977 Pedley and Guilleminault defined an atypical form of epilepsy, called episodic nocturnal wanderings, characterized by paroxysmal ambulation and bizarre behavioral manifestations during sleep. Even in the absence of recorded nocturnal seizures, the presence of interictal discharges on scalp encephalography and the good responsiveness to antiepileptic drugs induced these authors to consider such behaviors as epileptic manifestations during sleep.

Some years later Maselli and Oswald questioned the epileptic origin of these episodes, which were considered as night terrors with sleepwalking. More recently Plazzi et al found clear-cut epileptic discharges during typical episode of nocturnal wanderings in four subjects; they applied the term of epileptic nocturnal wanderings (ENWs) in order to define these kinds of seizures. Patients affected by ENWs showed other paroxysmal episodes of shorter duration resembling those previously described under the term of nocturnal paroxysmal dystonia and paroxysmal arousals. At present time paroxysmal arousals, nocturnal paroxysmal dystonia and ENWs are considered three clinical aspects of the same frontal lobe nocturnal epileptic syndrome. Invasive and semi-invasive recording with subdural grids and sphenoidal electrodes seems to confirm this hypothesis. Moreover, recently, using single-photon CT, Schindler et al found a significant hyperperfusion in the anterior part of the cingulate gyrus in a case of paroxysmal nocturnal dystonia.

We report the case of a drug-resistant epileptic patient affected by ENWs studied with stereotactically implanted intracerebral electrodes. Despite the suspected frontal origin of this kind of episodes, in our patient video-EEG features together with video-stero-EEG (SEE) data and the surgical outcome showed a temporal lobe origin of the seizures.

Case Report

The patient is a right-handed 39-year-old man with a negative family history for epilepsy and somnambulism. From age 6 the patient had nocturnal episodes while asleep during which he suddenly woke up, screamed, and had complex and wide movements involving both the upper and lower limbs. Sometimes he fell out of bed. In other episodes after showing complex motor behaviors, he got out of bed and walked around the room with violent movements, continuing to scream with a terrified expression. Lastly he went back to bed. His parents thought that he seemed to preserve consciousness throughout the seizure and that he was aware of it. These episodes lasted about one minute. Sometimes they were preceded by an aura characterized by fear, a gooseflesh sensation and/or visual hallucinations. Rarely he complained of subjective manifestations occurring during wakefulness, characterized by tachycardia and goose-flesh sensation associated with epigastric feelings and hyperventilation.

Frequency of the nocturnal attacks was 26 to 50 per month, sometimes with more than 10 attacks per night. At first they were considered as parasomnias, but later a diagnosis of epilepsy was made. However, no drugs were effective against these seizures, and the patient was sent to our center. MRI investigation and video-EEG monitoring with scalp and stereotactically introduced intracerebral electrodes were conducted. Neurologic examination and MRI were normal.

Interictal scalp EEGs showed paroxysmal epileptic abnormalities over the right frontotemporal region. Scalp video-EEG analysis showed detectable ictal patterns (flattening or fast activity) only during the few seconds preceding ictal clinical onset, then traces were rapidly obscured by movements artifacts. The EEG ictal onset was not well localized, showing a widespread temporal-frontal modification. On this basis an individualized SEE exploration was judged mandatory for precise surgical planning. Fourteen electrodes were positioned. As shown in figure 1, they explored the orbital gyrus, the cingulate gyrus, the middle frontal gyrus, the inferior frontal gyrus, the inferior parietal lobule, the hippocampus, the middle temporal gyrus, the amygdala, and the temporal pole.

During video SEE recording 4 stereotyped nocturnal episodes were recorded (at sleep-onset, during stage 2 sleep and during stage 3-4 sleep). During the attacks (Figure 1, upper part), the patient breathed in deeply, suddenly opened his eyes with a frightened facial expression and showed a brief pelvic thrusting. Then he began screaming, showing vio-
lent and wide legs movements. He sat forward and executed a clockwise rotation of the trunk and then left the bed and moved around the room continuing to scream with his arms stretched over his head. Extremely frightened and panting, he gripped the observer. Later he exhibited lip smacking and deglutition. Questioned at the end of the episodes, he could speak correctly and could remember what happened.

The SEEG ictal recordings (Figure 1, lower part) demonstrated a well-localized initial discharge always confined to the right temporal lobe structures: amygdala, hippocampus, para-hippocampus, and temporal pole. Such modifications preceded the first clinical sign in all the recorded seizures. The hyper-kinetic phase appeared when the discharge involved other temporal structures and the cingulate gyrus became the site of fast activity discharge.

The patient underwent a right temporal corticectomy including the anterior third of the temporal lobe, the amygdala, and the hippocampus.

After surgery the pharmacologic treatment was slightly reduced (carbamazepine from 1800 mg to 1600 mg per day; vigabatrin from 3500 mg to 3000 mg per day). Fifteen months postoperatively he is seizure free.

DISCUSSION

It has been reported that ENWs together with paroxysmal arousals and nocturnal paroxysmal dystonia constitute different manifestation of NFLE. This observation has not been confirmed by postoperative seizure-freedom criteria, and only in a few cases have recordings using sphenoidal electrodes or subdural grids been conducted. NFLE is characterized by different types of seizures often associated with inconclusive EEG ictal patterns so that the primary epileptogenic zone may not be clearly identified in the frontal regions in all patients. Moreover one out of four of the patients affected by ENWs reported by Plazzi et al had a aura characterized by a gastric rising sensation, a symptom that is more ascribable to temporal than to frontal lobe epilepsy.

Patients with temporal lobe epilepsy and complex partial seizures occurring predominantly or exclusively during sleep have been described. In these patients, seizures are less frequent and lack hyper-kinetic activity or complex motor automatisms as those observed in ENWs.

We do not have sufficient information to explain why patients affected by temporal lobe epilepsy rarely present with ENWs; however Pedley and Guilleminault in their first description of episodic nocturnal wanderings found interictal discharges in the right anterior temporal region in three out of six patients. These authors implicated the temporal lobe as the origin of the seizure activity in some of their patients, and our report seems to demonstrate the veracity of these findings.

Our patient showed the clinical features of ENWs with complex movements involving both the upper and lower limbs, followed by the typical agitated deambulatory pattern. The SEEG recording showed ictal discharges on the mesial temporal lobe structures starting some seconds before the clinical onset. The appearance of complex motor behaviors associated with intense fear coincided with the spread of the discharges to the other temporal structures and to the cingulate region. Reciprocal anatomic connections between the mesial and frontal areas as well as anterior cingulate regions have been proven to be strong; the cingulate region, via the hippocampus and para-hippocampus, constitutes a bridge between the temporal and the frontal lobes.

The cingulate gyrus has been shown to be hyperperfused both during the ictal phase of NPD and during episodes of sleepwalking; it seems to be a brain region highly involved, both primarily and secondarily, in the generation of complex motor behaviors during sleep. In a recent study it has been observed that in seizures characterized by intense fear with motor agitation, the epileptic discharge involves a
neuronal network constituted by the orbitoprefrontal, anterior cingulate, and temporal limbic cortices; the activation by stimulation of a single one of these regions did not induce the global clinical manifestation per se.15

Our findings, though limited to a single case, indicate that a clinical seizure pattern as that observed in ENWs could be elicited by discharges starting in the temporal structures with secondary involvement of the frontal lobe.

In most of subjects affected by ENWs, or more generally by NFLE, the identification of the epileptogenic zone within the frontal or temporal structures does not dramatically change the approach to patients because they usually respond well to drug therapy. However, drug-resistant patients affected by ENWs and nocturnal paroxysmal dystonia have been described,16 and in these cases the actual localization of the epileptogenic zone becomes crucial, considering that a surgical approach could be a resolutive remedy.

REFERENCES