INCE the seminal report of Pedley and Guilleminault, ENW has been thought to represent an atypical type of nocturnal complex partial seizure. Indeed, because of the bizarre complex motor pattern and the positive response to AEDs, ENW has been considered epileptic in nature. Moreover, although EEG recordings were at first inconclusive, the authors of more recent studies have shown that agitated somnambulent episodes similar to those previously described were associated with ictal epileptic discharges. Finally, it has been suggested that ENW may be part of the spectrum of so-called “nocturnal frontal lobe epilepsy,” also associated with paroxysmal arousal and nocturnal paroxysmal dystonia. Nocturnal frontal lobe epilepsy seems to be frequently cryptogenic, displaying a strong familial trait for parasomnias and epilepsy.

Until now, ENW has been poorly studied. Its epileptic nature has been a matter of debate. Furthermore, the frontal origin of ENW has been disputed. Finally, in a recent review of 100 cases involving NFLE, investigators showed that lesional ENW was rare and never associated with brain glioma. We report, for the first time in the literature, the occurrence of ENW in a patient harboring a right temporoinsular low-grade glioma. This ENW was resistant to AEDs before any surgery, whereas it completely resolved after tumor resection and the patient reported no recurrence during a follow-up period of 4.5 years. To the authors’ knowledge, this is the first report of ENW due to a glioma; the findings support the theory that ENW may represent an unusual type of lesional epilepsy that is surgically correctable. Moreover, a temporoinsular origin of ENW can now be considered.

KEY WORDS • nocturnal wandering • epilepsy • low-grade glioma • tumor resection

Case report

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Although controversial, episodic nocturnal wandering (ENW) is thought to be a rare and atypical form of nocturnal epilepsy, originating in the frontal lobe and responsive to antiepileptic drugs (AEDs). The authors report the case of a patient harboring a right temporoinsular low-grade glioma, who presented with a 3-year history of agitated somnambulent episodes resistant to AEDs. Interestingly, the ENW totally resolved after tumor resection and the patient reported no recurrence during a follow-up period of 4.5 years. To the authors’ knowledge, this is the first report of ENW due to a glioma; the findings support the theory that ENW may represent an unusual type of lesional epilepsy that is surgically correctable. Moreover, a temporoinsular origin of ENW can now be considered.

Case Report

HISTORY. This 38-year-old right-handed woman with no personal or family history of neurological disease, had epilepsy that had begun when she was 34 years of age. Initially, without any treatment, the mean seizure frequency reported by the patient was approximately one episode per day. Each seizure lasted between 2 and 3 minutes during sleep. The patient opened her eyes, jumped out of the bed, moved around, talked unintelligibly (without screaming), and acted in an agitated and, according to her husband, “mischievous” but nonviolent behavior.

The patient herself was not aware of her nocturnal motor activity but reported feeling tired during the daytime, after subjectively noting poor quality sleep; however, she maintained a normal socioprofessional life. Of note, she experi-

Abbreviations used in this paper: AED = antiepileptic drug; EEG = electroencephalography; ENW = episodic nocturnal wandering; MR = magnetic resonance; NFLE = nocturnal frontal lobe epilepsy.
experienced partial seizure during waking hours, with motor automatisms (scratching).

**Examination.** After suffering a generalized seizure 1 year after symptom onset, the patient was admitted to the hospital. Her neurological status was normal, and the Karnofsky Performance Scale score was 100. Magnetic resonance imaging revealed a right-sided paralimbic glioma involving the temporal pole, the anterior part of the temporomesial structures, and the insula (Fig. 1); according to the classification established by Yaşargil and Reeves, the lesion was a Type VB. Because of the likely diagnosis of low-grade glioma, due to the lack of enhancement on MR images, no surgery was proposed at that time, but the patient underwent regular follow-up MR imaging every 6 months.

A course of AEDs was instituted (valproate, carbamazepine, and topiramate). Despite a decrease in the mean seizure frequency (~ one per week), the patient continued to experience the same nocturnal symptoms during the following 3 years. Because of the presence of the tumor, only a simple scalp EEG study was performed, which showed interictal epileptic abnormalities such as a slow-wave activity with regular spikes over the right temporal region.

Taking into account a slight but regular increase in tumor volume documented on repeated MR imaging, we decided to undertake surgery in October 2000.

**Operation.** The resection of the glioma was performed using intraoperative electrical stimulation mapping to preserve the functional structures, especially the pyramidal pathways, according to the surgical methodology that we previously described concerning removal of tumors involving the insuloparalimbic system.

**Postoperative Course.** The postoperative course was uneventful. Neurological status was normal, the Karnofsky Performance Scale score remained at 100, and the patient returned to a normal life and full-time employment. Interestingly, no seizure has occurred since the surgery; AEDs were initially tapered down (only topiramate 50 mg per day) and were then stopped altogether during a follow-up period of 4.5 years.

Postoperative MR imaging demonstrated a subtotal resection, with residual tissue at 4 cm³ around the anterior perforated substance (Fig. 2). The histological examination of the pathological tissue showed a low-grade oligodendroglioma (World Health Organization Grade II) (Fig. 3).
No complementary treatment was given during the follow-up period. Nevertheless, a course of temozolomide was recently initiated when a tumor regrowth was demonstrated on control MR imaging; the patient’s neurological status, however, remained normal and no seizure had occurred.

Discussion

We have described the case of a patient presenting with agitated somnambulent episodes similar to those referred to as ENW. Moreover, the findings of interictal epileptic abnormalities on the simple scalp EEG, combined with the decrease in the mean number of episodes of seizure while receiving AED, supports the likely epileptic nature of this nocturnal wandering, reinforcing the current theory of ENW.12,14

There are several unique features in this case report. First, this is a lesional ENW, as these partial complex seizures were associated with a tumor. To our knowledge, this is the first observation of ENW due to a low-grade glioma. Indeed, in a recent review of the literature, ENW and associated neuroradiological abnormalities were found in only 14% of cases, and these were associated with vascular malformation, ischemic lesions, arachnoid cyst, cortical dysplasia, atrophy, and gliosis,13 but never with a tumor, in particular a glioma.

Second, our findings indicate that the resection of this lesion interrupted the ENW. Indeed, despite the reduction in seizure frequency from approximately 30 to five per month due to the AEDs, medical treatment did not provide complete relief of the epilepsy during a 3-year period. Conversely, immediately after the surgery, and during a follow up of more than 4 years, there was no seizure activity, despite the major reduction and final cessation of AEDs. Also noteworthy is that despite the extent of resection in eloquent areas, especially in the insula, there was no postsurgical permanent deficit, probably due to the brain plasticity mechanisms previously described.1 Consequently, because of the resolution of epilepsy, cessation of AEDs, and absence of postoperative adverse sequelae, the patient’s quality of life was improved by the surgery. It should be mentioned that one team recently described such a resolution of ENW following surgery, but the case involved a patient with nontumoral epilepsy.11,12

Third, ENW has traditionally been thought to have a frontal origin and is postulated to be part of a spectrum of NFLE.9,11,15 In our case, the findings seem to indicate that ENW could be generated at the level of the temporal and/or insular lobe. Indeed, the tumor corresponded to a right paralimbic low-grade glioma (Type VB according to the Yaşargil and Reeves classification17), which involved both the insula, the anterotemporal and mesiotemporal structures, but not the frontal lobe. Furthermore, scalp EEG monitoring showed interictal epileptic abnormalities exclusively over the right temporal lobe; however, because of the poor spatial resolution of this study, it was impossible to confirm (without the deeper-placed electrodes of stereo-EEG7) whether there was participation of the insular lobe, known to be potentially involved in some cases of temporal epilepsy.6 Finally, ENW resolved after the tumor was subtotally resected; the removal of paralimbic gliomas has been previously reported in the treatment of pharmacologically resistant epilepsy.2 In this way, our results are in accordance with two recent reports in which the findings support the...
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possible temporal lobe origin of ENW.\textsuperscript{10,11} Nevertheless, as mentioned in these reports, it is also possible that while the seizure origin is temporal, the hyperkinetic behavior is, rather, coincidental with the spreading of epileptic discharge to extratemporal regions and, in particular, to the frontal lobe (possibly via the insular lobe). If this hypothesis were applied to the patient in our case, it could be seen that the resection of the temporal tumor involved the seizure origin and also that the excision of the insula (even partial) may have disconnected the residual lesion from the frontal lobe.

Conclusions

In summary, the findings in the present case 1) show that ENW can be associated with paralimbic low-grade glioma, 2) provide further arguments in favor of the epileptic nature of somnambulistic episodes, 3) allow a better comprehension of the pathophysiology of ENW—potentially with the involvement of a larger network than the frontal lobe alone (that is, with the participation of the temporal and/or extratemporal lobes), and 4) may have possible implications in some cases of drug-resistant ENW, in which one third of cases\textsuperscript{9} are still considered candidates for surgical therapy.

References


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