

Epileptic Nocturnal Wanderings in A young Female: A rare Case Report and Review of the Literature

Alawi A. Al-Attas^{1*}, Attas A. Al-Attas, Maryam A. Al-Attas² and Saleh K. Swailem

¹Department of Neurology, King Saud Medical City, Riyadh, Saudi Arabia

²College of Medicine, Alfaisal University, Riyadh, Saudi Arabia

³King Fahad Specialist Hospital, Burydah, Saudi Arabia

Received: 20 April 2023

Accepted: 28 May 2023

*Corresponding author: alawiattas@yahoo.com

DOI 10.5001/omj.2024.37

Abstract

Sleep-related hyperkinetic seizures are a frequent symptom of frontal lobe epilepsy that occurs at night. Although temporal lobe-originating sleep-related seizures have been documented, they frequently lack hyperkinetic activity. Moreover, Episodic nocturnal wandering is rarely observed and is believed to represent an unusual form of nocturnal epilepsy that responds to anti-seizure medications. We present a 14-year-old right-handed girl with recurrent sleepwalking and wandering. Interictal electroencephalography revealed an epileptogenic focus in the left temporal lobe and intermittent slow wave activity originating from that region. During her nighttime wanderings, the patient exhibited unusually violent and nonviolent conduct, putting herself in danger of minor or serious injuries.

Keywords: Episodic nocturnal wandering, Epilepsy, Nocturnal frontal lobe epilepsy, Epileptic nocturnal wandering, Temporal lobe epilepsy.

Introduction

Sleep-related epileptic seizures and parasomnias are the two main categories of paroxysmal motor behaviors occurring during sleep.¹ In 1977, Pedley and Guilleminault defined episodic nocturnal wanderings (ENWs) as an unusual form of epilepsy marked by paroxysmal ambulation and abnormal behavioral and cognitive manifestations during sleep.² Even though no nocturnal seizures had been documented, the existence of interictal discharges on scalp electroencephalography (EEG) and a favorable response to anti-seizure drugs (ASMs) prompted the authors to suggest that ENWs represent an atypical form of epilepsy.³

The epileptic nature of these episodes was questioned by Maselli and Oswald, who interpreted them as night terrors with sleepwalking.^{4, 5} Years later, Plazzi et al. established the term epileptic nocturnal wanderings to describe similar seizures in four patients who displayed clear epileptic discharges during normal episodic overnight wanderings. In addition, they reported the coexistence of ENWs, nocturnal paroxysmal dystonia (NPD), and paroxysmal arousals (PAs), as well as similarities between these conditions. Therefore, they hypothesized that these three elements constitute a spectrum of sleep-related epilepsy.⁶

Even when seizures are a common symptom of intracranial mass lesions, ENWs have very rarely been observed in combination with obvious brain structure abnormalities.^{7, 8} In this report, we discuss the case of a young girl who had a rather unusual ENW presentation. The evidence in this case supports the idea that ENWs are probably actual epileptic seizures.

Case Report

14-year-old right-handed female began to have recurrent episodes of sleeping outside her bed at the age of 12. She occasionally strayed beyond her bedroom and did not remember the experiences. She sometimes responded instantly to verbal prompts, and at other times needed more time to respond. She did not exhibit oral or hand automatism, eye or facial twitching, jerky gestures, or sounds, or have a history of generalized tonic clonic seizures. The parents reported paroxysmal attacks while ambulating and doing sophisticated and structured motor activities. During these episodes, the patient remained silent and returned to bed if accompanied by another person. Even though she was aware that something had occurred throughout the night, she was unable to provide a detailed narrative of the incident the following day. Unfortunately, events were not recorded after her admission to the epilepsy monitoring unit (EMU).

The episodes were variable in frequency and clustering. Two and a half months prior to her most recent hospitalization, she began taking 500 milligrams of levetiracetam twice a day. During this time, her seizure frequency decreased, and she showed signs of improvement. She had approximately two minor injuries outside her bedroom, and despite taking her ASM, she was found outside her bed one or two nights per month. Stress was one of the aggravating factors for the events. She had no memory of the events. There are no other recognized risk factors in her past or family history. She does average work in school. Neurological examination was unremarkable. A routine laboratory test was also normal. MRI revealed an asymmetrical smaller left temporal lobe with ill-distinct gray–white matter differentiation. In addition, incomplete inversion of the left hippocampus was observed. (Fig. 1).

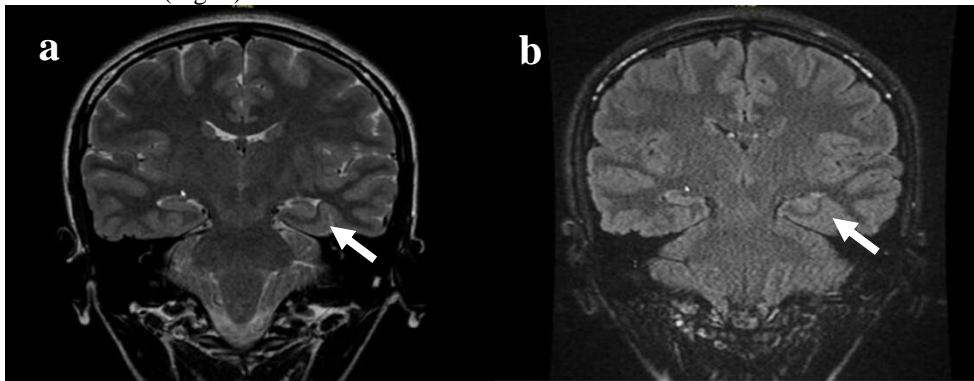


Figure 1: The MRI brain image (a-T2 and b-Flair sequences) shows an asymmetrical, smaller left temporal lobe with poorly distinct gray-white matter differentiation. In addition to the incomplete inversion of the left hippocampus (arrows).

The patient was readmitted to the EMU for further classification of her seizures and further treatment. Unfortunately, despite being off ASM, neither clinical nor electrographic seizures occurred during the patient's 12-day hospitalization. The neuropsychology and psychiatric teams evaluated her upon admission and excluded any psychiatric causes. The interictal EEG showed normal background activity and a focus of sharp wave activity in the left temporal lobe (Fig. 2). Due to undesirable side effects, such as agitation, levetiracetam was substituted after discharge. Lamotrigine was therefore given at a dose of 25 mg twice day for three days. The patient and her family were then instructed on how to progressively increase the dose to 200 mg twice daily. When she was assessed at the outpatient clinic three months later, she did not recollect any episodes of wandering.

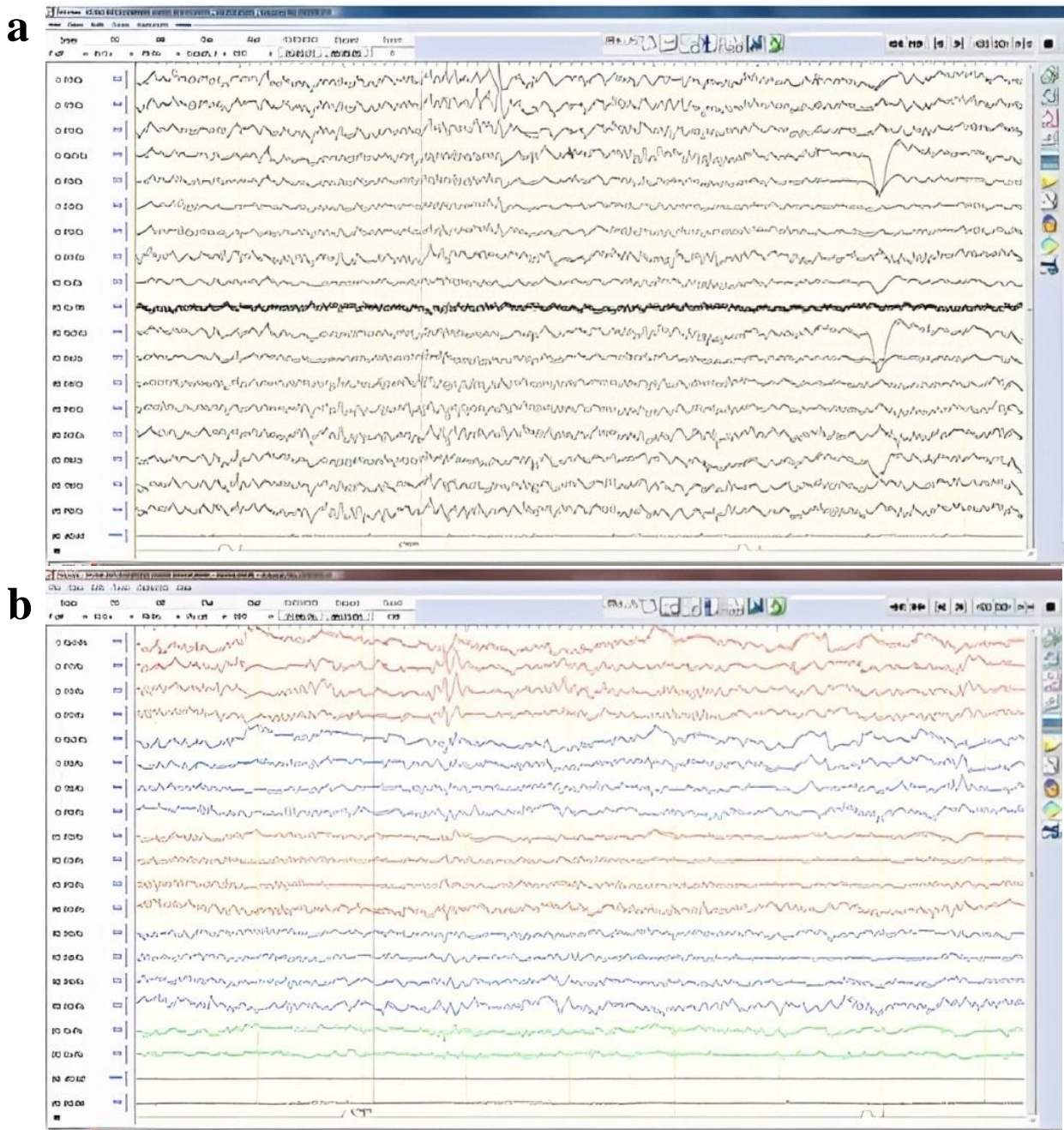


Figure 2: a-b depicts interictal epileptiform discharges, with sharp waves visible over the left temporal area in the a-CZ and b-bipolar montages.

Discussion

ENWs, paroxysmal arousals, and nocturnal paroxysmal dystonia are a few examples of the diverse nighttime frontal lobe epilepsy (NFLE) presentations that have purportedly been reported.³ The major epileptogenic zone in the frontal areas may not always be easily distinguished in some cases due to the presence of fuzzy EEG ictal patterns; furthermore, when NFLE was first identified in 1981, it was first referred to as nocturnal paroxysmal dystonia and thought to be a motor condition of sleep (NPD).⁶⁻⁹ It is challenging to identify NPD attacks from other non-epileptic nocturnal paroxysmal events, such as parasomnias, due to the atypical seizure semiology, onset during sleep, and frequently uninformative scalp EEG and brain MRI.⁸

The diagnosis of ENWs is complicated by the presence of frontal lobe hyperactivity or hypermotor activity during epileptic events, so it may be difficult to distinguish the primary epileptogenic zone in the frontal areas of

most patients.⁹ In the literature, there are occasionally clues that point to a potential temporal origin for sleep-related hyperkinetic seizures; for instance, when an anterior temporal cavernous angioma was removed from one of the 23 NPD patients described by Santiago et al., the patient's seizures ceased.¹⁰ The origin of epileptic nocturnal wanderings, a clinical symptom of NFLE, has also been suggested to be in the temporal lobe, according to a recent study.¹¹ Additionally, one in four of the patients who experienced the ENWs described by Plazzi et al. had an aura that was characterized by a feeling of the stomach rising, a symptom that is more consistent with temporal lobe epilepsy than frontal lobe epilepsy.^{6, 11} Focal impaired awareness seizures that mostly or exclusively occur during sleep have been described in patients with temporal lobe epilepsy. These individuals' seizures are less common and do not include hyperkinetic behavior or intricate motor automata such as those seen in ENWs.^{12,13}

In patients with generalized seizures, epileptic fugue, which was described during an absence or complex partial nonconvulsive status epilepticus, may occur in a postictal phase and frequently manifest as a frank confusional condition.¹⁴ Indeed, it has been described multiple times since 1956, when Gastaut et al. described an interesting case of a prolonged fugue state.¹⁵ Moreover, from 1887–1889, while serving as professor of neurology at the Salpêtrière Hospital in Paris, Jean-Martin Charcot gave a number of conversational case presentations on general neurology where he described a case with multiple fugue states, possibly of epileptic origin.¹⁶ Alejandro Jiménez-Genchi et al. reported the coexistence of epileptic nocturnal wanderings and an arachnoid cyst in a 15-year-old boy with a left temporal lobe arachnoid cyst.¹⁷ Furthermore, Huang et al. described episodic nocturnal wandering and complex visual hallucinations in a 25-year-old man with a left anterior temporal focus.¹

Our reported case is similar to Pedley et al., who described cases of episodic nocturnal wanderings that improved with ASMs in 1977, our patient demonstrated that ENWs have localized epilepsy that improves with ASMs.² According to Plazzi et al. and Provini et al., the frontal, temporal, or frontotemporal areas can be the anatomical locations of ENWs; indeed, diffuse interictal discharges (IIDs) and focal IIDs emanating from the right frontal lobe or left frontotemporal head regions are characteristic of the temporal or frontotemporal areas and their interictal EEG.^{6,7}

Identifying aberrant paroxysmal motor episodes during sleep presents a particular challenge for clinicians. On the one hand, these occurrences might be parasomnias, which are benign nonepileptic sleep disorders characterized as "unpleasant or undesirable behavioral or experiential phenomena that occur predominantly or exclusively during the sleep period." Parasomnias include sleepwalking and sleep terrors. On the other hand, these might be epileptic seizures, which would call for testing and care. In many cases, separating seizures from parasomnias using clinical history is rather simple. However, a specific type of epilepsy that is becoming more widely known presents a diagnostic dilemma.¹⁸

An unusual variety of nocturnal complex partial seizures, known as ENW, has been theorized to exist. Indeed, ENW has been characterized as epileptic in nature because of the peculiar, complex motor pattern and the good response to ASMs. In addition, although early EEG recordings were equivocal, more recent studies by other researchers have demonstrated that agitated somnambulant episodes like those previously described are connected to ictal epileptic discharges.¹⁹

Conclusion

Epileptic nocturnal wandering is one of the manifestations of sleep-related hypermotor epilepsy. It is essential to have a clinical suspicion of ENWs in the differential diagnosis of any paroxysmal motor behaviors occurring during sleep.

ENW is rare and thought to be an atypical form of nocturnal epilepsy that is responsive to ASMs. The present case provides more evidence that ENW is an uncommon kind of nocturnal focal impairment awareness seizure. In addition to other probable causes of nighttime wandering that are not caused by epilepsy, screening patients with nighttime wandering should consider the possibility of epilepsy.

Disclosure

The authors declare that they have no competing interests. Written consent was taken from the patient.

References

1. Huang Y, Seizure NC-, 1998 undefined. Episodic nocturnal wandering and complex visual hallucination. A case with long-term follow-up. *ElsevierPaperpile*, <https://www.sciencedirect.com/science/article/pii/S1059131198900115> (accessed 6 August 2022).
2. Pedley TA, Guillemainault C. Episodic nocturnal wanderings responsive to anticonvulsant drug therapy. *Ann Neurol* 1977; 2: 30–35.
3. Nobili L, Francione S, Cardinale F, et al. Epileptic Nocturnal Wanderings with a Temporal Lobe Origin: A Stereo-Electroen-cephalographic Study. *academic.oup.comPaperpile*; 25, <https://academic.oup.com/sleep/article-abstract/25/6/659/2750161> (2002, accessed 6 August 2022).
4. Maselli R, Rosenberg R, Sleep JS-, et al. Episodic nocturnal wanderings in non-epileptic young patients. *academic.oup.comPaperpile* 1988; 11: 156–161.
5. Oswald I. Episodic nocturnal wanderings. *Sleep* 1989; 12: 186–188.
6. Plazzi G, Tinuper P, Montagna P, et al. Epileptic nocturnal wanderings. *academic.oup.comPaperpile*, <https://academic.oup.com/sleep/article-abstract/18/9/749/2749736> (1995, accessed 6 August 2022).
7. Provini F, Plazzi G, Montagna P, et al. The wide clinical spectrum of nocturnal frontal lobe epilepsy. *ElsevierPaperpile*, <https://www.sciencedirect.com/science/article/pii/S1087079200901096> (accessed 6 August 2022).
8. Tinuper P, Bisulli F. From nocturnal frontal lobe epilepsy to Sleep-Related Hypermotor Epilepsy: A 35-year diagnostic challenge. *Seizure* 2017; 44: 87–92.
9. Kurahashi H, Hirose S. Autosomal Dominant Nocturnal Frontal Lobe Epilepsy. *GeneReviews®*, <https://www.ncbi.nlm.nih.gov/books/NBK1169/> (2018, accessed 19 August 2022).
10. Arroyo S, Santamaria J, Setoain JF, et al. Nocturnal paroxysmal dystonia related to a prerolandic dysplasia. *Epilepsy Res* 2001; 43: 1–9.
11. Nobili L, Cossu M, Mai R, et al. Sleep-related hyperkinetic seizures of temporal lobe origin. *Neurology* 2004; 62: 482–485.
12. Blair RDG. Temporal Lobe Epilepsy Semiology. *Epilepsy Res Treat* 2012; 2012: 1–10.
13. Bernasconi A, Andermann F, Cendes F, et al. Nocturnal temporal lobe epilepsy. *Neurology* 1998; 50: 1772–1777.
14. Das S, Ray BK, Dubey S. Temporal lobe epilepsy with nocturnal wandering leading to discovery of Moyamoya Angiopathy. *Acta Neurol Belg*. Epub ahead of print 2021. DOI: 10.1007/S13760-021-01830-Y.
15. Gastaut H, Roger J, neurologique AR-R, et al. The significance of certain epileptic fugues; concerning a clinical and electrical observation of temporal status epilepticus. *pubmed.ncbi.nlm.nih.govPaperpile*, <https://pubmed.ncbi.nlm.nih.gov/13351159/> (accessed 20 August 2022).
16. Goetz CG. Charcot at the Salpêtrière: ambulatory automatism. *Neurology* 1987; 37: 1084–1088.
17. Jiménez-Genchi A, Díaz-Galviz JL, García-Reyna JC, et al. Coexistence of Epileptic Nocturnal Wanderings and an Arachnoid Cyst. *J Clin Sleep Med* 2007; 3: 399.
18. Derry CP, Davey M, Johns M, et al. Distinguishing sleep disorders from seizures: diagnosing bumps in the night. *Arch Neurol* 2006; 63: 705–709.
19. Duffau H, Kujas M, Taillandier L. Episodic nocturnal wandering in a patient with epilepsy due to a right temporoinsular low-grade glioma: relief following resection. Case report. *J Neurosurg* 2006; 104: 436–439.