

CASE REPORTS

## Coexistence of Epileptic Nocturnal Wanderings and an Arachnoid Cyst

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Episodic nocturnal wanderings (ENWs) have rarely been associated with gross abnormalities of brain structures. We describe the case of a patient with ENWs in coexistence with an arachnoid cyst (AC). The patient was a 15-year-old boy who presented with nocturnal attacks characterized by complex motor behaviors. An MRI revealed a left temporal cyst and a SPECT Tc99 scan showed left temporal hypoperfusion and bilateral frontal hyperperfusion, more evident on the right side.

During an all-night polysomnographic recording with audiovisual monitoring, dystonic posture followed by sleepwalking-like behavior was documented. The sleepwalking-like behavior was preceded by a spike discharge over the left frontocentral region with contralateral projection

and secondary generalization during stage 2 sleep. Treatment with levetiracetam produced a striking remission of seizures. This supports a conservative management of an AC, considering that it may be an incidental finding. In epileptic patients, an AC may not necessarily be related to the location of the seizure focus.

**Keywords:** Episodic nocturnal wanderings, nocturnal frontal lobe epilepsy, seizures, arachnoid cyst.

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### INTRODUCTION

Episodic nocturnal wanderings (ENWs) were originally described as a condition characterized by recurrent episodes of bizarre behavior during sleep, interictal EEG abnormalities, and a good response to antiepileptic drugs.<sup>1</sup> Almost 20 years later, Plazzi et al.<sup>2</sup> documented ictal epileptic discharges over anterior brain regions during ENWs. They also reported both the coexistence of and similarities between ENWs, nocturnal paroxysmal dystonia (NPD), and paroxysmal arousals (PAs). They therefore suggested that these 3 entities represent a sleep related epileptic spectrum.<sup>3</sup>

Even when seizures are frequent manifestations of intracranial mass lesions, ENWs have rarely been found to coexist with evident brain structure abnormalities.<sup>3</sup> We describe the case of a patient with ENWs and a temporal lobe arachnoid cyst (AC).

### REPORT OF CASE

The patient was a 15-year-old boy with a family history that was negative for sleep, psychiatric, or epileptic disorders. His personal history revealed exposure to X-rays during the third tri-

mester of intrauterine life. His psychomotor development, school, and social functioning had been normal, and there was no previous history of sleep problems.

The first nocturnal episode occurred when he was 14 years old. While asleep on a bus during a family vacation trip, he suddenly woke up and walked to the front of the bus. He stood by the front windshield and stared for a few minutes. Then he went back to his seat and slept. This same behavior occurred again one hour later; both events were during the first half of the night. At about 03:00 of the following night, he suddenly got out of bed, struck the bed with his fists, and pulled out his hair. He returned to bed without replying to his mother's complaints. The patient did not remember any of the episodes and was surprised when told about them. During the third night, he got out of bed and threw himself onto the bed and accidentally hit his mother, who was sleeping in the same bed. His father held him down; the patient stared at him and went back to sleep.

At his home, similar episodes occurred 2 or 3 times per night. During the sleepwalking-like behaviors, he would walk into his parents' room, the bathroom, and kitchen. Frequently, he would also run and bang his head against the walls, chairs, and tables, while yelling obscene words or unintelligible sounds. Eventually, events began to occur during daytime naps as well. He reported no association between sleep behavior and dream content.

Four months later, he presented with urinary sphincter relaxation during an episode. He slipped and fell on the wet floor and suffered a jaw injury. Two weeks later, he experienced an absence seizure while watching TV. These attacks occurred 4 more times; some were associated with sphincter relaxation. A few days after, he was brought to our institution.

A psychiatric assessment identified diminished school performance, irritability, excessive appetite, hyperphagia, and a weight gain of 7.5 kg during a 2-month period. No psychiatric disorders were noted. A neurological examination and routine laboratory

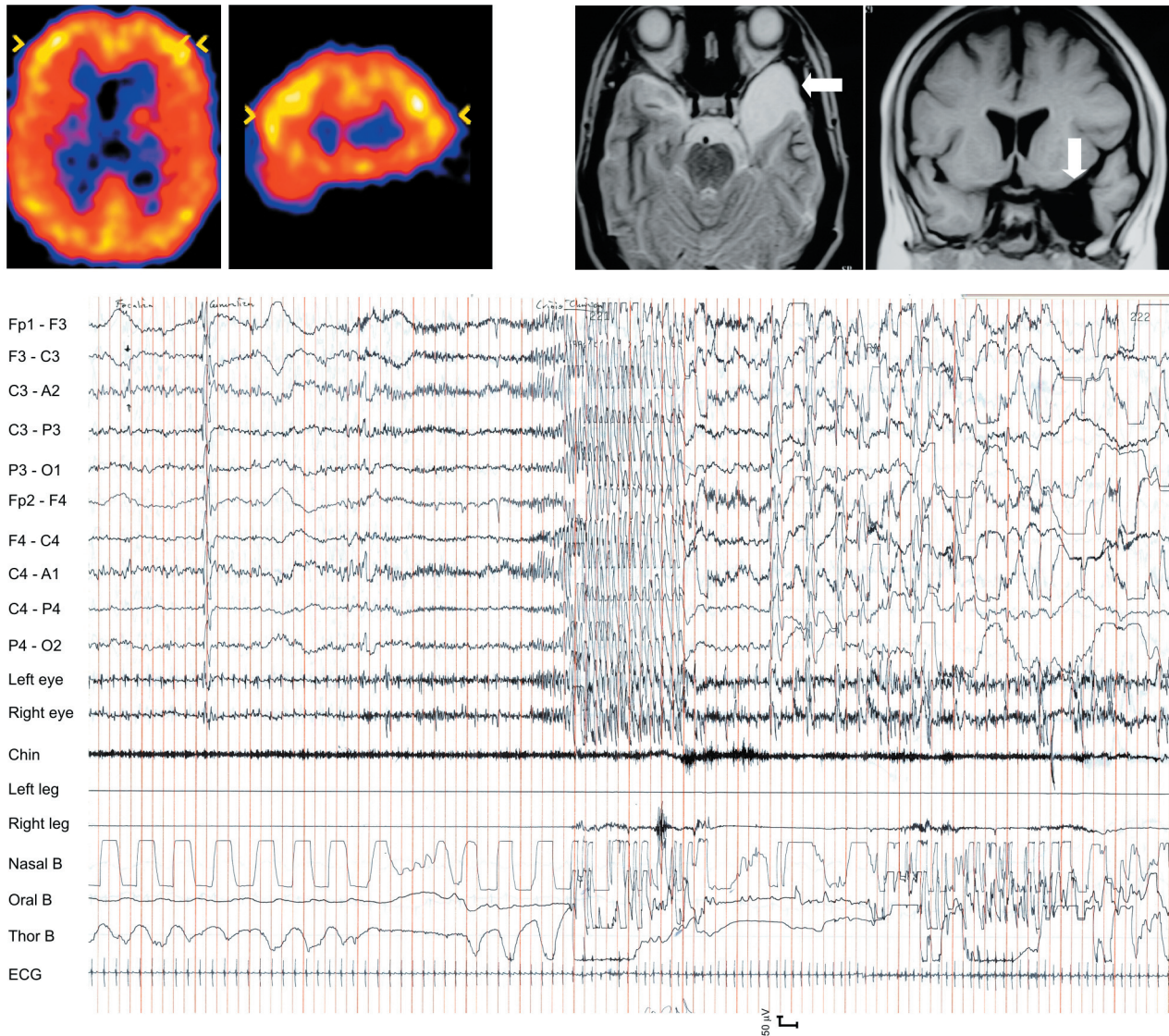
#### Disclosure Statement

This was not an industry supported study. Drs. Jiménez-Genchi, Díaz-Galviz, García-Reyna, and Ávila-Ordoñez have indicated no financial conflicts of interest.

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**Figure 1**—MRI images (upper right) show a large arachnoid cyst in the left temporal fossa. SPECT images (upper left) show a perfusion defect in cyst site (coronal slice) and hyperperfusion in bilateral frontal cortex (transverse slice). Before the beginning of an ENW (bottom), EEG shows a spike discharge over the left frontocentral region with contralateral projection and secondary generalization after 2 seconds. The clinical attack begins during an arousal. Tachycardia and irregular breathing are also present.

studies were normal. An interictal EEG showed paroxysmal discharges over the right frontotemporal region. Magnetic resonance imaging revealed a left temporal cyst (Fig 1). An interictal 99mTc-ECD single photon emission computed tomographic (SPECT) scan showed a perfusion defect in the cyst site and bilateral frontal hyperperfusion, more evident on the right side (Figure 1).

During an all-night polysomnographic (PSG) recording with audiovisual monitoring, the patient had several paroxysmal episodes characterized by sudden tonic hyperextension of the right hemibody. This was followed by the extension and the abduction of the right limb with the flexion of his fingers, as if he was trying to cover himself with a sheet. This motor behavior occurred 8 times. In one episode, the motor behavior was different: he suddenly sat up, put his legs out of the bed, touched the head leads, and then tried to get out of bed but was stopped by the PSG technician. This last behavior was preceded by a spike discharge over the left frontocentral region with contralateral projection and secondary generalization during stage 2 sleep (Figure 1).

Treatment with magnesium valproate, 1000 mg/day produced only a slight decrease of seizure frequency. Three months later,

levetiracetam, 1000 mg/day was added, and there was a striking remission of attacks. For over a year, he has presented with neither nocturnal nor diurnal seizures. Irritability, hyperphagia, and excessive appetite have disappeared, while his weight and school performance have normalized. An EEG performed one year later was normal.

## DISCUSSION

Arachnoid cysts are non-tumorous fluid collections ascribed to abnormalities in embryonic development of the arachnoid membrane.<sup>4</sup> They usually present in children, usually boys, and their most common symptoms include raised intracranial pressure, focal neurological deficits, headaches, and seizures. However, ACs may also be found in asymptomatic subjects.<sup>5</sup>

The prevalence of ACs in patients with epilepsy is low (1.9 %).<sup>6</sup> Likewise, brain lesions are rarely found in patients with ENWs.<sup>3</sup> To our knowledge, the coexistence of ENWs and ACs has not been reported.

Little is known about the anatomic and neurophysiological relations between ACs and epileptic activity. It has been reported

that ACs may be incidental findings in epileptic patients and are not necessarily related to the location of the seizure focus.<sup>6</sup> Our observations in this case are consistent with what has been reported. The seizures documented during the PSG had an origin in the frontal lobe, which also showed hyperperfusion in the interictal SPECT scan. It should be noted that the frontal lobe is the most frequent origin of ENWs, NPD, and PAs.<sup>3</sup>

Because arousal disorders may begin in adolescence, they must be considered in the differential diagnosis of the patient's nocturnal behavior.<sup>7</sup> Based on clinical data, there are 3 characteristics which support an epileptic etiology in this case: a) the high frequency of episodes, particularly during the same night; b) the appearance of events during daytime naps; and, c) the appearance of an absence seizure during wakefulness. The characteristics of the events during polysomnographic recording with audiovisual monitoring are also consistent with an epileptic origin because the attacks were stereotypic, repetitive, and involved dystonic movements. In contrast, episodes of typical parasomnias are low in frequency, and the nocturnal behavior is not stereotypic and repetitive.

Both treatment resistant epilepsy and ACs, isolated or comorbid, may be an indication for surgery. However, no superiority of surgical treatment over the conservative approach has been reported.<sup>8</sup> Our findings, even though they are limited to one case, support a conservative management, where levetiracetam may have a useful role.

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