Nocturnal Epilepsy Masquerading as a Behavioral Problem in Childhood

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Nocturnal epilepsy has been described as a clinical syndrome since the early 1950s. It has been reported most frequently in the European literature and receives scant attention in current American pediatric texts. Two cases are described that presented as behavioral problems in childhood but whose subsequent evaluation and response to therapy support the diagnosis of nocturnal epilepsy. Clues in the history and the failure of behavioral intervention are important in suspecting the syndrome. Physical examination and waking electroencephalographic findings are normal in most cases. Electroencephalography performed during sleep provides positive evidence of nocturnal epilepsy in the majority of cases.

Nocturnal epilepsy, which is also known as hypnic, morpheic, or nonphasic epilepsy,[1] appears to have had little recognition as a clinical diagnosis. Textbooks give it brief mention or omit it entirely. For example, in the 10th edition of the *Nelson Textbook of Pediatrics* by Vaughan and McKay,[2] the following description is found: "A grand mal seizure may occur at night (nocturnal epilepsy) without the patient being aware of it. A bitten tongue or lip, headache, blood on his pillow or a bed wet with urine may be the only clue."

Nocturnal epilepsy is an important diagnosis that, like many lesser known illnesses, may be detected only through its specific inclusion in the differential diagnosis. It may occur as a truly nocturnal neurologic disturbance, which may have minimal, overt manifestations of the epileptic state during the awake hours. This paper will describe and discuss two cases of confirmed nocturnal epilepsy that were initially diagnosed and treated as behavioral problems. The diagnoses were missed in these patients for periods of 18 months and two years, respectively. Ultimately, the diagnoses resulted from abnormal findings being detected following an electroencephalogram performed during sleep. A dramatic resolution of symptoms followed treatment with phenytoin and helped provide therapeutic confirmation of the diagnosis.

Case 1

A mother brought her nine-year-old daughter to the Family Practice Center with "a behavioral problem." The child was inattentive at school, irritable and uncooperative with her teachers, and frequently fell asleep during classes. The symptoms were of 18 months' duration and began shortly after the family had moved from another state. The child had also begun to complain, episodically, of headache on awakening. When she awakened with a headache, she would remain irritable

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and sleepy for the remainder of the day. Two or three such episodes were occurring each month.

Six months after the onset of symptoms, the patient had been seen by a neurologist. There had been no increase in the frequency or severity of the problem, but its persistence was alarming the parents. A complete neurologic examination followed by skull x-ray examination and an awake electroencephalogram had revealed no abnormality. It was suggested that the recent family move was the precipitating factor, and the child's symptoms and behavior were attributed to stress resulting from difficulty in adjusting to her relocation.

During the year prior to being brought to the Family Practice Center, the child's attacks had insidiously increased in frequency. They were occurring two to three times each week. Concurrently, the child's schoolwork had deteriorated to the point that she was in danger of failing a grade.

Past medical and family history were unremarkable; physical examination revealed a well-nourished, alert child of normal developmental age with no detectable abnormalities. On the basis of the history, and because previous neurologic studies had been negative, an electroencephalogram during sleep was the only initial investigation performed. Interpretation revealed "a local disturbance in the left temporal lobe suggesting the possibility of an underlying seizure disorder." Following review of the case with a pediatric neurologist, a diagnosis of nocturnal epilepsy was made and treatment was commenced with phenytoin. There was immediate resolution of the problem. The headaches and associated behavior did not recur. The child became alert and cooperative; with extra teaching she has been able to remain in her appropriate grade. One year later she remains on phenytoin and free of symptoms.

Case 2

A five-year-old boy was brought to the Family Practice Center by his grandmother, with a two-year history of "severe behavioral problems." The pattern of abnormal behavior was erratic. On a bad day the child was difficult to arouse in the morning and would sleep until early afternoon if left undisturbed. When forced to get out of bed on these occasions, the boy would complain of headache, and he would be subject to temper tantrums. At times he would become uncontrollably destructive; he would break furniture and physically hurt his sisters. No system of reward or punishment had been found to moderate these violent outbursts. This behavior occurred two to three times each week and could be predicted from the child's attitude on morning awakening. On other days the boy's behavior would be normal and appropriate for a child of his age.

He was a second child, who had begun to misbehave shortly after the birth of a sister. Both parents had been in their teens at the time of marriage, and three children occurring in as many years following their union had been a severe stress. Family dysfunction had resulted, and the maternal grandmother had assumed care of the children. The patient's behavior had become so unmanageable that the grandmother had allowed his placement in a foster home. The parents had supported this decision. In the foster home the child had been receiving psychiatric counseling at weekly intervals during the two years prior to being seen at the Family Practice Center. Further deterioration in his behavior had been observed over this two-year interval. The boy had recently been allowed a trial stay in his grandmother's home, and it was during this period that he was brought to the Family Practice Center.

Behavioral and developmental milestones were reported as being achieved at appropriate ages.

Physical examination revealed a normal, well-behaved child in a good state of nutrition, with normal growth and no dysmorphic features. Careful neurologic and physical examination revealed no abnormalities. Skull x-ray examination was normal. The awake electroencephalogram was reported as normal; the sleep electroencephalographic report noted "both an abnormal diffuse and local pattern, the disturbances being present in the left posterior quadrant strongly suggestive of an underlying seizure disorder." In consultation with a pediatric neurologist, the patient began a course of phenytoin therapy. An immediate improvement in behavior was noted; the temper tantrums ceased, and psychiatric counseling was discontinued.

The boy returned to his grandmother's care. His mother now has joined her children in her parent's home and is gradually resuming the care of her children. The patient now exhibits normal, manageable behavior for a five-year-old child.

Discussion

The reported prevalence of childhood epilepsy has varied from 2 to 8 cases per 1000 population.[3] Nocturnal epilepsy, or epilepsy occurring exclusively during sleep, is uncommon at all ages.[4] Seizure discharge on electroencephalogram is much more common in sleep than in the waking state.[5] Oswald[6] revealed much of the present knowledge of sleep, and Janz[7] reviewed the information covering epilepsy and the sleep-waking cycle. Lennox-Buchthal,[8] in a study of febrile convulsions, found a close relationship between the convulsion and the sleep-waking cycle in that over one half of the attacks occurred while either going to sleep or on awakening. This pattern has also been confirmed in primary, generalized grand mal epilepsy[9] and in benign local epilepsy; 51 percent of the cases had their attacks exclusively during sleep.[10]

Ambrosetto and Gobbi[11] were able to show spike discharges activated in rapid eye movement sleep and slow wave sleep, the actual epileptic attack occurring on awakening while the patient was still drowsy. Tassinari et al[12] described a very rare type of epilepsy occurring during slow wave sleep, involving children between the ages of 4 and 10 years, who exhibit infrequent nocturnal seizures, diurnal absences, prepsychotic behavioral disorders, and deterioration in intellectual capacity and social performance. Despite treatment, this condition shows slow, but never complete, remission.

The two cases described here fit all the criteria of nocturnal epilepsy: no sign of diurnal convulsions, intermittent "postictal behavior," a positive sleep electroencephalogram, and prompt improvement with treatment.

Of course, it should not be inferred that all behavioral problems of childhood fall into the category of an underlying epileptic disorder, nor should it be assumed, however, that all behavioral problems of childhood result from stress or parental mismanagement.

As these two cases reveal, if postictal symptoms become persistent, and the intensity of abnormal behavior occurs at a level that disturbs the quality of a child's life, suspicion should begin. The parent's chronic frustration at "mobilizing" the child on these mornings, the unusual intensity of the subsequent temper tantrums, the same-day drowsiness and inattention in the classroom, all provided grounds for suspicion in these two cases.

Persistent, recurrent headache in the patient and chronic parental frustration in aiding the child may be additional clues. Marital dysfunction may accompany or result from the child's illness. Inability on the part of parents and teachers to modify the child's behavior with reward or punishment and the ability of parents to predict the child's response constitute major, presumptive evidence compatible with a diagnosis of nocturnal epilepsy.

The electroencephalogram is much more likely to support a diagnosis of nocturnal epilepsy when conducted during sleep than when performed during the awake state.[13] One study utilizing the awake electroencephalogram had negative findings in 70 percent of the cases that ultimately proved to be due to nocturnal epilepsy.[14]

Whenever doubt exists, a short-term treatment with phenytoin may provide the diagnosis through rapid, symptomatic improvement. As revealed through the cases described here, the potential benefits of phenytoin therapy in this disorder far exceed its known side effects for such a therapeutic trial.

References

1. Passonant P, Latour H, Cadilhac J: Hypnic epilepsy. Ann Med Psychol 2:526, 1959
2. Vaughan VC, McKay RJ (eds): Nelson Textbook of Pediatrics, ed 10. Philadelphia, WB Saunders, 1975
3. Berg BO: Prognosis of childhood epilepsy—Another look. N Engl J Med 306:14, 1982
4. Gibberd FB, Bateson MC: Sleep epilepsy: Its pattern and prognosis. Br Med J 2:403, 1974
5. Gibbs FA, Gibbs EL: Atlas of Electroencephalography, vol 2: Epilepsy. Cambridge, Mass, Addison-Wesley, 1952
6. Oswald I: Sleep (ed 3). London, Penguin, 1974
7. Janz D: Epilepsy and the sleep-waking cycle. In Vinken PJ, Bruyn GW (eds): Handbook of Clinical Neurology, vol 15: The Epilepsies. Amsterdam, North-Holland, 1974, pp 457-490
8. Lennox-Buchthal MA: Febrile convulsions: A reappraisal. Electroencephalogr Clin Neurophysiol (suppl) 32: 38, 1973
9. O'Donohue NV: Epilepsies of Childhood. Woburn, Mass, Butterworth, 1979
10. Beaussart M: Benign epilepsy of childhood with rolandic (centro-temporal) paroxysmal foci: A clinical entity. Study of 221 cases. Epilepsia 13:795, 1972
11. Ambrosetto G, Gobbi G: Benign epilepsy of childhood with rolandic spikes, or a lesion? EEG during a seizure. Epilepsia 16:793, 1975
12. Tassinari CA, Terzano G, Capocchi G, et al: Epileptic seizures during sleep in children. In Penry JK (ed): Epilepsy, The Eighth International Symposium. New York, Raven Press, 1977, pp 345-354
13. Janz D: The grand mal epilepsies and the sleeping-waking cycle. Epilepsia 3:69, 1962
14. Christian W: Bioelectrical characteristics of forms of epileptic diseases bound to the periods of the day. Dtsch Z Nervenheilk 181:413, 1960