Four-Year-Old Girl with Abnormal EEG on Routine Overnight Polysomnogram for Snoring and Behavioral Issues

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A 4-year 10-month-old female was referred to the pediatric sleep clinic for intermittent snoring with periods of observed gasping. She underwent adenoidectomy at 3 years 11 months, after which nasal congestion, snoring, and increased nighttime awakenings again recurred. Parents reported neck hyperextension, frequent mouth breathing, and daytime sleepiness, in addition to restless sleep. She frequently came to sleep in parents' bed, and mother noted intermittent episodes of hyperextension of arms and legs with associated tremors but not tonic-clonic activity.

She was diagnosed with autism spectrum disorder after a developmental evaluation for delayed social skills. She was born full term via C-section without complications due to breech presentation. She has no history of seizures or other neurological disorders nor a family history of seizures. However family history was significant for snoring in parents and half-brother with learning disability and tic disorder. Prior trials of antihistamines resulted in daytime sedation, and steroidal nasal sprays were associated with behavior issues.

Examination showed a cooperative quiet girl with height and weight appropriate for age. She was noted to have frequent mouth breathing. On oropharyngeal exam, tonsils were graded 2-3+. Moderate nasal congestion without obvious turbinate hypertrophy was noted. Cardiorespiratory and neurologic exam were normal.

Patient underwent an overnight polysomnogram. Evaluation of the respiratory component noted mild supine dependent sleep apnea, with an RDI of 4.6, and supine RDI of 8.4 with lowest oxygen saturation of 96%. On the accompanying EEG, the following was noted (see **Figure 1**).

Figure 1— 30-second epochs prior to and after sleep onset

(A) 30-second epoch showing EEG abnormality in the right central lead prior to sleep onset. (B) 30-second epoch soon after sleep onset with increased frequency of spike discharges in right central region during NREM sleep. LEOG AND REOG, left and respectively right outer canthus electro-oculography electrodes; F3-M2, C3-M2 and O1-M2, left frontal, central and respectively, occipital electroencephalography electrodes; F4-M1, C4-M1 and O2-M1, right frontal, central and respectively, occipital electroencephalography electrodes; Chin EMG, submental electromyography signal; ECG II, one standard electrocardiogram lead; TFLOW, the airflow via nasal/oral thermocouples; PFLOW, the airflow via nasal air pressure transducer; CHEST and ABDO, chest and respectively abdominal walls motion via inductive plethysmography; SpO2, the pulse oximetry by finger probe; PLETH, plethysmographic waveform; POSITION, S = Supine

QUESTION: What do the findings on the EEG (Figure 1) represent?

ANSWER: Recurrent centro-temporal spikes seen on EEG during wakefulness but increased in NREM sleep are suggestive of benign rolandic epilepsy (BRE) or benign epilepsy of childhood with centrotemporal spikes (BECTS).

DISCUSSION

Epileptiform discharges are found in 1% to 2% of pediatric polysomnograms, and incidence may be higher in those with sleep disordered breathing.^{1,4} Additionally, children with autism spectrum disorders are at an increased risk of seizures with epileptiform abnormalities reported on EEG in 10% to 50% of cases.² Centrotemporal spikes seen on EEG in children between 3 and 13 years of age is typical of BRE. Peak age of onset for BRE is 5-8 years with a male predominance. Seizures in BRE present with unilateral facial or oropharyngeal muscle involvement, hypersalivation, speech arrest and sometimes tonic-clonic activity. They are usually brief, lasting for 1-2 minutes often occurring during sleep or just before awakening, but may become secondarily generalized.⁵ Remission is usually within 2-4 years of onset and often by adolescence. The overall total number of seizures is also small, with the majority of patients having less than 10 seizures; 10% to 20% have only a single seizure.⁵ Some patients may not have seizures, and this case study emphasizes the need for EEG findings to be correlated with clinical history.

EEG evaluation is the primary tool for diagnosis with unilateral or bilateral centro-temporal spikes, seen during wakefulness and markedly accentuated by NREM sleep. The EEG evaluation should include an overnight EEG, prolonged daytime EEG to capture sleep, or a polysomnogram with additional bilateral EEG electrodes for a definitive diagnosis. Only those with cognitive impairment, very frequent epileptiform discharges on EEG, and history of generalized seizures are generally treated.⁵ Such patients should undergo neurological evaluation.

CLINICAL PEARLS

1. Epileptiform discharges are found in 1% to 2% of pediatric polysomnograms and are seen at a markedly higher frequency in autism spectrum disorders.

2. Focal centro-temporal spike wave discharges on the EEG are suggestive of benign rolandic epilepsy.
3. Neurological evaluation is recommended in patients with non-sleep related nocturnal paroxysmal events.
4. Polysomnography with an extended EEG montage along with video recording is recommended for evaluation of paroxysmal nighttime awakenings that may be seizure related.³

CITATION

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