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Childhood paroxysmal nonepileptic events

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ABSTRACT

We aimed to determine the types and clinical characteristics of paroxysmal nonepileptic events (PNEs) in children. During a 13-year period, 765 patients underwent long-term video-EEG monitoring, and 95 (12.4%) of them were identified to have PNEs. The most common diagnoses were conversion disorder, parasomnias, staring spells, movement disorders, and hypnic jerks. Paroxysmal nonepileptic events originated from physiologic or organic (43.2%) and psychogenic (56.8%) causes. Mean delay in diagnosis was 3.1 years. Mean ages at diagnosis were 8.8 and 13.8 years in physiologic or organic and psychogenic groups, respectively. A marked female predominance was seen in the psychogenic group, whereas males slightly predominated in the physiologic or organic group. In the physiologic or organic group, events were less frequent, longer in duration, and commonly manifested as subtle motor activity, whereas subtle and prominent motor activities were encountered equally in both groups. Concomitant epilepsy was present in 10.5% of the patients. Differences in clinical characteristics may be helpful in differentiating physiologic or organic PNEs in children from psychogenic PNEs.

1. Introduction

Paroxysmal nonepileptic events (PNEs) are episodic changes in behavior, sensation, or consciousness that resemble epileptic seizures but are not associated with abnormal ictal cerebral electrophysiological discharges [1]. They may occur in all age groups. In most instances, the clinical history leads to the correct diagnosis, and ancillary testing serves as confirmation; in a subgroup of cases, however, an accurate diagnosis is one of the clinical problems faced by practitioners [2].

While psychogenic seizures and cardiac events comprise the majority of PNEs in adults, besides these disorders, a wide variety of physiologic and organic disorders including parasomnias (confusional arousals, sleep walking, sleep terrors, and nightmares), sleep-related movement disorders (periodic limb movements in sleep, nocturnal leg cramps, and rhythmic movement disorders), narcolepsy, benign paroxysmal nocturnal events (hypnic jerks and benign sleep myoclonus of infancy), breath-holding spells, Sandifer syndrome, and behavioral events can mimic seizures in children. There are no somatic causes for psychogenic PNEs, rather, they are somatic manifestations of psychologic distress [3]. The onset of psychogenic PNEs is typically in the adolescence or young adulthood period [4]. In a group of patients diagnosed with psychogenic PNEs, most patients experienced the first episode between 10 and 19 years; however, correct diagnosis was established between 20 and 40 years [4,5].

Long-term video-EEG monitoring offers simultaneous assessment of both clinical events and cerebral electrical activity, so it has a great importance in differentiating epileptic seizures from nonepileptic seizures, as well as in seizure classification and presurgical evaluation [1,6–9]. Paroxysmal nonepileptic events are among the most common causes of treatment-refractory spells, and up to 43% of patients seen at pediatric epilepsy referral centers may have a paroxysmal nonepileptic disorder [2,10,11]. In a tertiary epilepsy center, the epilepsy diagnosis was disproved in 30% of children referred without any doubts about the epilepsy diagnosis [2]. The misdiagnosis of epilepsy has important consequences, including unnecessary exposure to antiepileptic drugs and unnecessary exposure to invasive interventions such as intubation and even to invasive therapeutic modalities such as vagal nerve stimulation implantation [12]. Early recognition and appropriate treatment of nonepileptic seizures can prevent significant iatrogenic harm and may result in a better outcome.

Data concerning the relative frequency and phenomenology of childhood PNEs by etiology are limited [6,7]. While staring episodes and unresponsiveness have been reported as more common manifestations in patients with physiologic or organic PNEs, prominent motor activity was more common in adolescents who mostly had psychogenic PNEs [6,13].

In this study, we aimed to evaluate the demographic features of children with PNEs and the nature, relative frequency, and clinical manifestations of the events documented in our pediatric long-term video-EEG monitoring unit during a 13-year period and to determine

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whether these features differ in children with PNEs that originated from organic or physiologic and psychogenic causes.

2. Materials and methods

Video-EEG reports of 765 children who were admitted to the pediatric video-EEG monitoring unit at Gazi University Medical Faculty between October 1998 and May 2012 were reviewed. Pediatric Neurology Department of Gazi University Medical Faculty is a tertiary epilepsy referral center for children. An Institutional Ethical Committee approved the study. Children who had a clinical diagnosis of paroxysmal nonepileptic events (PNEs) were identified. Paroxysmal nonepileptic events were defined as paroxysmal changes in behavior, not associated with a seizure pattern on scalp EEG recordings. Only patients who had at least one of their typical episodes documented on video-EEG were included into the analysis. For children admitted more than once, only data from one admission were included into the study. Neonates were monitored in neonatal intensive care unit and excluded because of the specific characteristics of this age group. Children with clear clinical diagnosis of paroxysmal disorders such as syncope, migraine, breath-holding spells, tics, shuddering attacks, and parasomnias were not monitored and excluded from the study. Demographic, clinical, and video-EEG data including duration of symptoms prior to diagnosis, frequency and duration of events, previous and/or current use of antiepileptic medication, medical history in regard to premature birth, perinatal asphyxia, febrile convulsions, perinatal infections, and/or trauma, and semiology of events in regard to the type of motor activity were gathered. Prominent motor activity was defined as abrupt paroxysmal changes in motor activity that resembled an epileptic convulsion, such as focal or complex motor activity, generalized jerking, and generalized tremor. On the other hand, subtle motor activity was defined as paroxysmal benign events such as episodes of staring or daydreaming, generalized limpness, sensory symptoms, bursts of crying or shouting, or clearly stereotypical episodes with walking, running, rocking, or repetitive movements. Besides psychogenic seizures, a wide variety of physiologic and organic conditions can cause paroxysmal spells mimicking epileptic seizures [6,14]. Based on the etiology of the recorded events, we divided children with PNEs into psychogenic and physiologic or organic groups. Psychogenic PNEs included conversion reaction which was defined as abrupt paroxysmal changes in behavior or consciousness resembling an epileptic seizure which could be associated with secondary gain [3] and events originated from a psychiatric disorder. Physiologic or organic events included paroxysmal behavioral disorders which originated from an organic or physiologic etiology, such as sleep-related disorders, movement disorders, syncope, and staring spells.

Ictal and interictal EEG patterns were recorded by using a 32-channel digital video-EEG system (Telefactor Beehive System, Telefactor, Philadelphia, PA) which included automatic spike and seizure detection modules. Scalp electrodes were placed according to the International 10–20 System with additional bilateral inferior temporal chains. Entire interictal EEG patterns recorded during wakefulness and sleep and all clinical events identified by parents or caregivers or children were reviewed by one or more trained pediatricians on a daily basis for the presence of epileptiform discharges. Senior epileptologists (A.S., T.H.), together with one or more pediatricians, then reviewed each EEG and video segments of these episodes and determined whether they represented an epileptic or nonepileptic event. The diagnosis of the nonepileptic event was determined on the basis of its semiology and clinical characteristics and with the absence of any ictal epileptiform abnormalities on the EEG. The diagnosis of PNEs was made only if family members verified that the monitored events matched the patients' typical spells. A concomitant diagnosis of epilepsy was made if an epileptic seizure episode was also captured during the video-EEG monitoring and there were epileptiform abnormalities on the EEG.

Patients were monitored for 1 to 7 days, depending on whether sufficient numbers of the typical events occurred. Antiepileptic drugs were tapered or discontinued during the monitoring period as necessary in patients who were on treatment. In patients suspected of having psychogenic seizures, when typical episodes were not observed, an attempt was made to induce the event by verbal suggestion, hyperventilation, and/or the injection of 1- to 2-ml saline intravenously after obtaining parental consent. In 95 patients, the typical spells with no accompanying epileptic discharges on EEG recording were captured; in the remaining 27 patients, PNEs were diagnosed on the basis of clinical history and prolonged EEG recordings that did not show any epileptiform discharges.

Statistical analysis was performed using SPSS 15.0 for Windows. The Student t test, Pearson's χ^2 test, and Fisher's exact test were used to compare demographic and clinical characteristics between the two groups and considered statistically significant if the p values were <0.05.

3. Results

From October 1998 to May 2012, 765 children underwent long-term video-EEG monitoring, and 122 (15.9%) of them had a clinical diagnosis of PNEs. Ninety-five (12.4%) patients had at least one of their typical events during the monitoring and were included in the analysis. The remaining 27 patients whose habitual events were not captured during the monitoring were excluded from the study.

Fifty-four (56.8%) patients were determined to have psychogenic PNEs, and except for 2 children who were diagnosed with hyperactivity disorder and impulse control disorder, all patients in this group had conversion reactions (54.2%). The remaining 41 (43.2%) patients had physiologic or organic PNEs, and the more frequent diagnoses in this group included parasomnias (12.6%), staring spells (11.6%), movement disorders (10.5%), and hypnic jerks (6.3%). The distribution of PNE disorders diagnosed by video-EEG monitoring is presented in Table 1.

Demographic and clinical characteristics of patients are shown in Table 2. Of 95 patients, 54 (56.8%) were females and 41 (43.2%) were males. The percentage of females (66.7%) was higher in the psychogenic group, whereas there was a slightly higher proportion of males (56.1%) in the physiologic or organic group ($p=0.027$). Male predominance was more marked in patients with parasomnias, followed by patients with hypnic jerks and movement disorders (Fig. 1). When all patients were analyzed as a single group, the mean ages at the time of onset of symptoms and at the time of diagnosis were 8.5 ± 4.5 years and 11.6 ± 4.4 years, respectively. Mean age at onset was significantly lower in patients with physiologic or organic PNEs when compared with patients with psychogenic PNEs (4.9 ± 3.3 years in the physiologic or organic group, 8.8 ± 4.5 years in the psychogenic group, $p<0.001$). Similarly, mean age at diagnosis was significantly lower in patients

Table 1
Types of paroxysmal nonepileptic event disorders by diagnosis and age at onset for each disorder.

Disorders	Number	Mean age at onset ± SD yr (range)
Psychogenic events	**54 (56.8)** [a]	**13.79 ± 2.81**
Conversion disorder	52 (54.7)	14.05 ± 2.49 (5–18)
Psychiatric disorder	2 (2.1)	7.00 ± 2.82 (5–9)
Organic or physiologic events	**41 (43.2)**	**8.82 ± 4.53**
Parasomnia	12 (12.6)	8.41 ± 4.07 (3–15)
Staring	11 (11.6)	7.27 ± 5.23 (2–16)
Movement disorder	10 (10.5)	11.00 ± 4.49 (4–17)
Hypnic jerk	6 (6.3)	8.33 ± 4.17 (3–15)
Syncope	1 (1.1)	8.00
Sleep-related breathing disorder	1 (1.1)	13.00
Total	95 (100)	11.65 ± 4.39

SD, standard deviation; yr, years.
[a] Values are numbers (%).