Paroxysmal Nonepileptic Events in Children and Adolescents

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ABSTRACT. Objective. Paroxysmal nonepileptic events (PNEs) are frequently encountered in children and adolescents; however, there is little information concerning the relative frequency of various types of these disorders. We report our experience with PNEs in a group of children and adolescents who underwent prolonged video-electroencephalographic monitoring.

Methods. During a 6-year period, 883 patients were monitored in the Pediatric Epilepsy Monitoring Unit and 134 patients (15.2%) were documented to have PNEs on the basis of a typical spell recorded during monitoring. Their hospital charts were reviewed and videotapes of these events were analyzed.

Results. Patients were divided into 3 age groups: 1) the Infant, Toddler, and Preschool Group (2 months–5 years) that comprised 26 patients. The most common diagnoses were stereotyped movements, hypnic jerks, parasomnias, and Sandifer syndrome. Concomitant epilepsy was present in 12 patients (46%). 2) The School-Age Group (5–12 years) consisted of 61 patients. The most frequent diagnoses were conversion disorder (psychogenic seizures), inattention or daydreaming, stereotyped movements, hypnic jerks, and paroxysmal movement disorders. Fifteen patients (25%) had concomitant epilepsy. 3) The Adolescent Group (12–18 years) consisted of 48 patients, of whom 40 patients (83%) were diagnosed with conversion disorder. Nine patients (19%) had concomitant epilepsy.

Conclusions. In our patients with PNEs, conversion disorder was seen in children >5 years old and its frequency increased with age, becoming the most common type of PNEs among adolescents. In adolescents, conversion disorder was more common in females, whereas males predominated in the school-aged group. Concomitant epilepsy with nonepileptic events occurred in all 3 age groups to a varying extent. Pediatrics 2002;110(4).

MATERIALS AND METHODS

Between January 1989 and December 1995, 883 patients <18 years old underwent video-EEG monitoring in the PEMU at the Cleveland Clinic Foundation. Among these patients, 199 (22.5%) were discharged with a final diagnosis of PNEs. PNEs were defined as paroxysmal changes in behavior, not associated with a seizure pattern on scalp EEG recordings. Patients were monitored from 1 to 5 days, depending on the number of events. If no spontaneous events occurred during this period, an attempt was sometimes made to induce an episode by suggestion. In 134 patients, we succeeded in capturing their typical spells; in the remaining 65 patients, PNEs were diagnosed on the basis of clinical history and prolonged EEG recordings over several days that did not show any epileptiform discharges. Events with symptomatology consistent with epileptic seizures that did not show EEG changes (such as auras or mesial frontal lobe seizures) were excluded. We reviewed the hospital charts and videotapes of 134 patients in whom at least 1 typical event was captured during continuous video-EEG monitoring. This included 1 patient with factitious disorder (Munchausen Syndrome by proxy) in whom no events were captured and the diagnosis was reached after a detailed clinical and social work evaluation. Patients with documented PNEs accounted for 15.2% of all patients monitored in our PEMU. All recorded events were confirmed with a parent or guardian to be the child’s typical spells. The patient was interviewed during and after their episodes by technologists, nurses or physicians in the PEMU using verbal, visual, or tactile stimuli to get their attention. A patient was judged to be unresponsive when all such measures failed to get any response. Older children were also asked to recall test words and items presented to them during recorded events. In patients strongly suspected of having psychogenic seizures, when no episodes were observed over 4 or more days of recording, we attempted to induce such an episode after obtaining parental consent. These methods included sleep deprivation, verbal suggestion, hyperventilation, photic stimulation, and/or the injection of saline intravenously. A concomitant diagnosis of epilepsy was made if an epileptic seizure was also captured during the video-EEG monitoring or the description of other events was judged to be highly suspicious for an epileptic seizure given the presence of interictal epileptiform discharges on EEG. All EEG and video segments of these episodes were analyzed by one of the authors (M.C.) and also reviewed with one of the senior authors (P.K. and E.W.).

Based on the pathophysiology of the recorded events, we divided the PNEs into 2 groups: psychiatric disorders and those that
were either physiologic or organic. Psychiatric disorders were diagnosed after a thorough evaluation by a child psychiatrist at our institution, using the criteria listed in the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition. Psychogenic seizures are subdivided into: a) somatoform or conversion disorders; b) dissociative disorders; c) anxiety disorders; d) disorders with psychotic symptoms; e) factitious disorder and malingering; and f) reinforced behavior patterns.

RESULTS

The 134 children and adolescents with PNEs were grouped by age: the Infant, Toddler, and Preschool Group (2 months old–5 years; mean: 3.17 years) 26 patients (17 males and 9 females); the School-Age Group (age 5–12 years; mean: 6.16 years) 61 patients (35 males, 26 females); and the Adolescent Group (age 12–18 years; mean: 15.5 years) 48 patients (17 males, 31 females).

Physiologic or organic disorders were seen in 66 patients (Table 1). The more frequent diagnoses included inattention/daydreaming, stereotyped movements, hypnic jerks, and parasomnias. All of the patients’ episodes of inattention or daydreaming were interruptible by tactile or verbal stimulation with the exception of a 2-year-old boy, whose staring episodes could not be aborted. In this child, the staring episodes lasted 5 to 30 seconds, occurred up to 20 times a day, and showed no EEG seizure pattern. This child was otherwise completely normal.

Psychiatric disorders were seen in 69 patients, including 1 patient in the Infant, Toddler, and Preschool Group (a 2-year-old boy with factitious disorder), 26 patients were in the school age group and 42 patients in the adolescent group. In the School-Age Group, 4 boys were felt to have episodic dyscontrol syndrome/intermittent explosive disorder, and 22 patients had conversion disorder (8 females: 14 males). In the Adolescent Group, 2 patients, both girls, were diagnosed with panic attacks and 40 with conversion disorder (26 females: 14 males). Our youngest patient with conversion disorder was a 5-year-old boy. The distribution of the psychiatric versus physiologic or organic disorders according to age is shown in Fig 1. Conversion disorder was more frequent among males in the School-Age Group (14 males: 8 females; \( P = .20 \)), which was opposite of that found in the Adolescent Group (26 females: 14 males; \( P = .06 \)). Although the proportion of male to females within each age category was not statistically significant, a significant difference emerged when the 2 groups were compared with each other, using the \( \chi^2 \) test (\( P < .03 \); Fig 2).

Based on clinical manifestations, patients with conversion disorder group could be divided into 2 groups: a) unresponsive events, during which the patient became unresponsive with reduction or the absence of spontaneous movements (lasting as long as 40 minutes in one instance) or b) motor events, in which they exhibited motor phenomena, often consisting of bizarre, irregular, jerking or thrashing movements of the extremities, not typical of any of the known types of epileptic seizures. In some patients, the level of responsiveness would change abruptly during the course of the event. In the School-Age Group, there were 14 patients with unresponsiveness as the main manifestation while 8 patients had predominantly motor events. In the Adolescent Group, 26 patients had unresponsive events, 17 had motor events, and 3 patients had both types of episodes. These differences in symptoms between the School-Age and Adolescent Groups were not statistically significant.

A concomitant disorder such as epilepsy, developmental delay, or both was found in 59 patients (Table 2). This occurred in 21 (34%) of 62 patients with conversion disorder. In 14 (67%) of these 21 patients, the concomitant diagnosis had been present before the discovery of conversion disorder (this included 11 patients with epilepsy or 17.7% of those with conversion disorder). These 11 patients with epilepsy and conversion disorder accounted for 1.5% of our patients with epilepsy studied in the PEMU.

In the physiologic or organic disorders group, a concomitant diagnosis was present in all 14 patients with inattention or daydreaming. Concomitant epilepsy was present in 2, developmental delay with abnormal neurologic examination in 3, developmental delay with epilepsy in 5, and attention deficit disorder in 4 patients. Ten (91%) of 11 patients with hypnic jerks had a concomitant diagnosis: epilepsy in 3, developmental delay with abnormal neurologic examination in 1, developmental delay with epilepsy in 5, and attention deficit disorder in 1. Among the 41 patients with organic disorders, 16 (39%) patients had a concomitant diagnosis: epilepsy in 3, developmental delay in 11, and epilepsy with developmental delay in 2.

The duration of symptoms before reaching the correct diagnosis averaged 1.35 years (range: 3 weeks–4 years). At the time of admission into the PEMU, 88 patients were on antiepileptic drugs (AEDs). Forty-one patients were discharged from the hospital on an AEDs; 36 patients were proven to have concomitant epilepsy on the basis of recorded seizures, whereas 5 patients had interictal epileptiform discharges on their EEG (3 patients with focal and 2 with generalized sharp waves).

Follow-up information was available for only 35 patients. Among the conversion disorder group, 20 patients were seen at a median time of 8.35 months.

<table>
<thead>
<tr>
<th>TABLE 1. Physiologic and Organic PNE Disorders by Diagnosis</th>
<th>Age</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>2 Months–5 Years</td>
</tr>
<tr>
<td>Inattention/daydreaming</td>
<td>1</td>
</tr>
<tr>
<td>Hypnic jerks</td>
<td>4</td>
</tr>
<tr>
<td>Stereotyped movements</td>
<td>5</td>
</tr>
<tr>
<td>Parasomnias</td>
<td>5</td>
</tr>
<tr>
<td>Movement disorders</td>
<td>0</td>
</tr>
<tr>
<td>Gastroesophageal reflux</td>
<td>4</td>
</tr>
<tr>
<td>Nonepileptic myoclonus</td>
<td>2</td>
</tr>
<tr>
<td>Apneas</td>
<td>2</td>
</tr>
<tr>
<td>Shuddering attacks</td>
<td>1</td>
</tr>
<tr>
<td>Alternating hemiplegia</td>
<td>1</td>
</tr>
<tr>
<td>Migraine</td>
<td>0</td>
</tr>
<tr>
<td>Hyperventilation attacks</td>
<td>0</td>
</tr>
<tr>
<td>Syncope</td>
<td>0</td>
</tr>
<tr>
<td>Total</td>
<td>25</td>
</tr>
</tbody>
</table>
(range: 3–36 months) and 65% of them were free of events. Among patients in the physiologic/organic disorders group, only 15 were followed up at a mean of 5.1 month (range: 3–12 months); only 20% were free of their events.

Although only 10 patients with parasomnias underwent EEG-video monitoring in the PEMU, during the same period, another 60 children and adolescents with parasomnias were evaluated as outpatients in the Pediatric Sleep Disorders Clinic, some of whom were also evaluated in the sleep laboratory. All 10 patients monitored in the PEMU were found to have nonrapid eye movement parasomnias (night terrors, [4]; sleepwalking, [4]; and confusional arousals, [2]).

**DISCUSSION**

PNEs are quite commonly encountered in infants, young children, and adolescents. In a substantial proportion of cases, a careful history and examination will elucidate their nature. However, in other cases, it is necessary to differentiate PNEs from epileptic seizures by prolonged EEG-video monitoring. Bye et al.\(^5\) recently published their experience with PNEs in a group of children undergoing video-EEG monitoring. They encountered PNEs in 285 (43%) of 666 patients, outnumbering children with epileptic seizures (40%) and those in whom events were not captured (17%). This somewhat high percentage of PNEs may reflect a referral bias. In our patient population, PNEs accounted for 23% of infants, children, and adolescents admitted to the PEMU for video-EEG monitoring. In 68% of diagnosed cases, we succeeded in documenting the typical episodes of the patient. In the remaining patients, a persistently normal EEG over several days of monitoring while off antiepileptic medications strengthened the clinical suspicion that the episodes were likely PNEs. Our study was limited to patients referred for EEG-video monitoring and therefore is not a true indication of the actual incidence of PNEs in the general population.

Approximately half the documented PNEs were physiologic or organic in nature, and the remaining half comprised psychiatric disorders of which conversion disorder (psychogenic seizures) was the most common entity, especially among adolescents.

TABLE 2. Concomitant Diagnoses With PNE Disorders

<table>
<thead>
<tr>
<th></th>
<th>Hyposics Jerks/Inattention N = 25</th>
<th>Organic Disorders N = 41</th>
<th>Conversion Disorders N = 62</th>
<th>Psychiatric or Other Diagnoses N = 7</th>
</tr>
</thead>
<tbody>
<tr>
<td>Epilepsy</td>
<td>5</td>
<td>3</td>
<td>11</td>
<td>0</td>
</tr>
<tr>
<td>Developmental delay</td>
<td>4</td>
<td>11</td>
<td>7</td>
<td>0</td>
</tr>
<tr>
<td>Developmental delay and epilepsy</td>
<td>10</td>
<td>2</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td>Total number with concomitant diagnosis</td>
<td>19 (76%)</td>
<td>16 (39%)</td>
<td>21 (34%)</td>
<td>0</td>
</tr>
</tbody>
</table>
In the School-Age Group, conversion disorder was less frequent than physiologic/organic disorders, accounting for 36.6% of PNEs in that age group; the youngest patient with conversion disorder was 5 years old. The only psychiatric entity encountered below that age range was factitious disorder, which typically involves an infant or very young child. Careful documentation of this disorder by EEG-video monitoring is important to the welfare of the child so that appropriate psychiatric and social work intervention can be provided promptly. In the absence of clear evidence to suggest this diagnosis, one should evaluate the child and family carefully to avoid making false accusations that could lead to a parent losing custody of the child.18–21 As recommended by Gates,17 we do not use the term pseudo-seizures.1,30 A key feature of psychogenic seizures is that patients with seizures that are not epileptic in origin do have a true illness, whereas the word pseudo is pejorative, carrying the connotation of false or deceptive. The terms nonepileptic event and nonepileptic seizure have been used interchangeably in the literature and refer to both psychiatric and nonpsychiatric conditions.

Except for 2 adolescent females with panic attacks, conversion disorder accounted for nearly all of the psychiatric diagnoses among adolescents, two-thirds of the patients being girls. This female predominance has been noted in other reports of psychogenic seizures among adolescents22–24 as well as in adult patients.25–27 However, in the School-Age Group, boys outnumbered girls by a ratio of 7 to 4. The reason for this age-related difference is unknown. Additional studies are needed to confirm this hypothesis. In addition to conversion disorder, episodic dyscontrol syndrome or intermittent explosive disorder was also encountered in this age group; all 4 children were boys. This agrees with previously reported literature.16

Leis et al28 noted that unresponsiveness without motor manifestations is the most common feature of psychogenic seizures; they studied 47 patients ages 4 to 51 years, most of whom were adults. Kramer22,29 found that unresponsive events were more common in school-age children compared with adolescents but this difference was not statistically significant. Kramer also found that psychogenic seizures with motor activity were significantly more common among adolescents.29 We found a similar proportion of unresponsive versus motor events, 63% and 60% respectively, in both school-aged and adolescent groups. Some adolescents exhibited both motor and unresponsive types of events. The motor phenomena of psychogenic seizures have been described in detail in the literature, and we also found them to comprise irregular movements that could switch from one extremity to another, side-to-side head movements, pelvic thrusting, and abrupt cessation. The level of unresponsiveness could also change abruptly during or immediately after the motor activity. We found the icteral and postictal interview of the patient to be very helpful in documenting "psychogenic unresponsiveness," a key feature of psychogenic seizures.1,30

Although concomitant epilepsy has been reported in 15% to 60% of patients with psychogenic seizures,11,12,22,31 recent data suggests that this number is perhaps closer to 10%.30–32 We found that concomitant epilepsy, developmental delay, or both occurred in 18%, 11%, and 5%, respectively, of children and adolescents with psychogenic seizures. Conversely, only 11 (1.5%) of 746 children with epilepsy had psychogenic seizures, compared with 20% of adults with chronic epilepsy reported to have psychogenic seizures.34–36 This may reflect the effects of chronic epilepsy over a number of years, higher incidence of abuse, maladaptive behaviors, or lack of access to effective psychotherapy. Documentation of both psychogenic and epileptic seizures is essential for optimal management of these patients.

We found that 35% of our patients with PNEs had been started on AEDs unnecessarily. In the series of Leis et al,28 75% of patients had been placed on AED therapy (6 also received emergent treatment for status epilepticus). It appears after being given a diagnosis of epilepsy, some individuals become overly dependent on family support and attention. In some of these cases, psychogenic seizures become more apparent once epileptic seizures have been brought under better control.

Among physiologic and organic disorders, daydreaming or inattention episodes comprised the largest category. Such episodes of lapses in attention, daydreaming, drowsiness, or stereotyped behavior occurring in a mentally retarded child,37,38 may raise the concern among parents, teachers, or physicians that the child could be having absence seizures. More than half of our patients with episodes of staring or inattention (14/25, or 56%) had developmental delay, which could make it harder to make this distinction on the basis of clinical description alone. Other entities included stereotypies, which are more common in neurologically impaired children, movement disorders such as tics, myoclonus, paroxysmal dystonia or choreoathetosis, shuddering attacks, and gastroesophageal reflux.39,40 Parasomnias were underrepresented in our patient sample because they were usually evaluated in the sleep laboratory or diagnosed on clinical grounds alone. Because of the retrospective nature of this study, only limited follow-up was available for most of these children with PNEs.

We find inpatient video-EEG monitoring to be much more useful than ambulatory EEG monitoring because it provides videotape documentation of the events and allows an interview of the patient during events—this is particularly important in psychogenic seizures. The monitoring should be long enough to capture at least 1 typical event with good quality video and EEG; at our institution, this ranges from 1 to 7 days. Patients with >1 type of seizure or event should be carefully evaluated for concomitant epilepsy. Limitations of ambulatory EEG monitoring include: a limited number of channels available for recording, contamination of the EEG by artifacts attributable to loose leads, movement and muscle activity, and a lack of objective documentation of the events. However, in seizures that are reliably associated with seizure patterns detectable on scalp EEG,
ambulatory recordings can quantify the number of seizures in a given time period, eg, absences or secondarily generalized seizures.

CONCLUSION

Video-EEG monitoring is essential to making an accurate diagnosis of PNEs, reassuring the family that all care has been taken to exclude epileptic seizures, and helping to keep open the lines of communication between the patient’s family and physician. In some patients who have epileptic and psychogenic seizures, the treatment plan needs to address both disorders. The gender predominance of males among younger children with psychogenic seizures differs from that seen in adolescent and adult patients.

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http://www.pediatrics.org/cgi/content/full/110/4/e46
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