

## Psychogenic Nonepileptic Seizures in Children and Adolescents with Epilepsy

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### ABSTRACT

Psychogenic nonepileptic seizures (PNES) are defined by episodes of abnormal movement, sensations, or cognitive experiences similar to epileptic seizures. These events, however, are not related to abnormal electrical brain discharges and are thought to be caused by a psychological process. Children appear to carry a lower risk for PNES compared to adults and yet this diagnosis may be present in up to 10% of the pediatric patients. The main features of PNES in such age group is discussed.

**Key words:** epilepsy, nonepileptic seizures, nonepileptic psychogenic seizures, nonepileptic psychogenic seizures in children.

### RESUMO

*Crises não-epilépticas psicogênicas em crianças e adolescentes com epilepsia*

Crises não-epilépticas psicogênicas (CNEP) são definidas como episódios de movimentos anormais, sensações ou experiências cognitivas similares a crises epilépticas. Estes eventos, contudo, não são relacionados a descargas elétricas cerebrais anormais e parecem estar associadas a processos de natureza psicológica. CNEP são menos frequentes em crianças quando comparados a adultos, porém ainda assim este diagnóstico pode ser observado em 10% da população de pacientes pediátricos. As principais características de CNEP nestes grupo etária são discutidas nesta revisão.

**Unitermos:** epilepsia, crises não-epilépticas, crises não-epilépticas psicogênicas, crises não-epilépticas psicogênicas em crianças.

### INTRODUCTION

Psychogenic nonepileptic seizures (PNES) are episodes of abnormal movement, sensations, or cognitive experiences similar to epileptic seizures.<sup>1-4</sup> However, these manifestations are not associated with abnormal electrical brain discharges and are caused by a psychological process.<sup>5</sup>

It is thought that children carry lower risk for PNES than adults,<sup>6</sup> but this may represent the underdiagnosis of this condition in childhood, on account of as there are only few studies on this specific topic. According to

Holmes et al.,<sup>5</sup> PNES are diagnosed in about only 10% of pediatric patients.

The person with epilepsy has an estimated \$ 100,000 lifetime cost for diagnostic testing, procedures, and medications. Repeated work-ups and treatments for what is mistakenly thought to be epilepsy are estimated to incur \$ 100 to 900 million per year in medical services.<sup>7</sup>

Therefore, PNES is an important issue in treating adults and children with intractable epilepsy. PNES in children as it is the case in adults impair quality of life and lead to iatrogenic consequences. The work of Lanctman

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et al.,<sup>8</sup> showed that there was an important impact on patient's quality of life which was evident when the seizures were present as compared to the seizure-free periods.

For these reasons, one must be aware of its prevalence, clinical features and some of the risk factors in order to recognize these events earlier and provide adequate treatment.

## PREVALENCE

The occurrence of PNES in adults referred to tertiary care centers ranges from 17 to 30%.<sup>6</sup> In a study by Kotagal et al.,<sup>9</sup> the frequency of paroxysmal nonepileptic events, including psychogenic events, in children and adolescents was 15.2%.

According to Ritter and Kotagal,<sup>10</sup> of 842 children evaluated in the pediatric epilepsy monitoring unit at Cleveland Clinic from 1989 to 1995, 199 (23.6%) were diagnosed to have non-epileptic events; these were recorded by Video-EEG in 168 (20%) patients. Of these patients, 35 (20.8%) had epilepsy in addition to their non-epileptic events.

Five to twenty percent of children and adolescents with epilepsy have PNES.<sup>6</sup> Vincentiis et al.<sup>11</sup> studying children and adolescents with epilepsy reported a higher prevalence of approximately 30.3%. It corroborates studies in adults that show that PNES are more frequent in people with epilepsy.

## CLINICAL FEATURES

According to the literature, some of the main features for the clinical diagnosis of PNES are: female predominance, older age, events with a peculiar semiology and longer duration, normal EEG, and normal neuro-imaging studies.<sup>12</sup>

Clinical features may help or suggest the diagnosis but are not a gold-standard method for the diagnosis especially in children who also have epilepsy. The diagnosis based on clinical history is difficult and tricky, leading most clinicians to request a video-EEG monitoring. However, some clinical findings are helpful in order to guide the diagnosis and for this reason they will be discussed.

### 1 Gender

As for gender distribution, the female predominance (66-99% of cases) is controversial, being reported in some pediatric population studies,<sup>6,8</sup> but not in others.<sup>11,13</sup> According to Pakalnis and Paolicchi,<sup>13</sup> 11 (69%) of 16 were boys, suggesting that contrary to findings in the adult population, PNES is commonly seen in boys.

These differences may be partially explained by the inclusion of younger children (preschoolers and lower-school groups). In early ages, the gender predominance does not seem as relevant as in adolescents and adults.

### 2 Age

Studies in adults demonstrate two peaks of incidence in the occurrence of PNES: 19-22 years and 25-35 years; occurrence before 5 years is reported to be rare.<sup>6</sup>

Wyllie et al.<sup>14,15</sup> demonstrated that PNES may occur in young children between 8 and 10 years of age. According to these authors, younger children, with nonepileptic paroxysmal behavior, frequently present mannerisms that may be mistakenly diagnosed as epilepsy or psychogenic seizures, such as parasomnias, hypoventilation attacks, breathholding spells, syncopal events, and movement disorders.

On the other hand, frank episodes of psychogenic unresponsiveness or "convulsions" are frequently observed later in life, by the end of the first decade.<sup>6,8,16-18</sup>

Although it is uncommon, some patients at the age of 6 years or younger had true psychogenic seizures. The earlier age of onset seems to more evident when one analyses children with personal or family history of epilepsy.<sup>11</sup>

### 3 Neurologic history

A record of a previous neurologic disorder is frequently reported in children with PNES, however some common maladies such as head trauma are not as common as in adults.

In a study of adolescents with PNES, there was a positive neurologic history in 21% of these patients.<sup>8</sup> A history of meningitis and arachnoid cyst were the most frequently encountered neurologic diseases in these patients history.

Head trauma was reported as a common findings in the study of Pakalnis and Paolicchi,<sup>13</sup> seven (44%) of the 16 patients with PNES had an antecedent history of head injury prior to the development of these episodes.

In the work of Lancman et al.,<sup>8</sup> 21 patients (48.8%) were taking anticonvulsants. The presence of a neurologic history frequently misguides the diagnosis, emphasizing that the presence of a previous neurologic history does not exclude the occurrence of PNES.

### 4 Family history

A family history of epilepsy was found in 37.6% of adults with PNES.<sup>19</sup>

Similar rates have been documented in children with PNES. In a study carried out with 43 patients exhibiting psychogenic seizures with onset before the age of 16 years, with a mean age at seizure onset of 12.4 years (range, 5 to 16 years), showed family history of epilepsy in 15 cases (34.9%).<sup>8</sup>

It may be postulated that the possibility to witness an event may represent a risk factor for PNES in children.<sup>8</sup>

Family history of epilepsy or a personal history of epilepsy,<sup>20</sup> may be an important modeling effect in presentation of this type of conversion symptom complex as part of their underlying psychiatric illness. Also, observation of other family members with epilepsy and the sick role may have provided some type of secondary gain and avoidance of family and school obligations.<sup>21</sup>

## 5 Physical and psychological abuse

Another important risk factor in adults, namely, physical or sexual abuse, was not documented as frequently in pediatric series. Psychological abuse, characterized by direct verbal aggression perpetrated by relatives or other close/familiar individuals, was documented in few patients.

On the other hand, Lancman et al. found a significant personal and family distress in most of the cases.<sup>8</sup> The presence of inadequate family setting, characterized by a stressful environment at home not directly related to the patient but affecting the patient's life according to his or her own perception, was considered a stress factor and was quite frequent, corroborating the notion that children are extremely susceptible to environmental influences, mainly family setting as a stress factor.<sup>11</sup>

In the work of Wyllie et al.,<sup>21</sup> 15 patients (44%) had severe family stressors including recent parental divorce, parental discord, or death of a close family member and only 2 patients (6%) had a history of physical abuse.

Previous studies that investigated sexual abuse in an older population subset of primarily adolescent and adult female patients in contrast to the group of school-age children.<sup>21,22</sup> History of sexual or physical abuse, which has been a well-recognized risk factor in development of psychogenic seizures in adults, are not frequently reported in children.<sup>23</sup>

## 6 Psychiatric disorders

The psychophysiological mechanisms that cause PNES are controversial,<sup>6</sup> and therefore, the issue is how PNES should be categorized in psychiatric terms.

There seems to be a consensus about the high comorbidity of mood disorders in children with PNES.<sup>20,21,24,25</sup> Mood disorders, mainly depression, are significantly more frequent than others. This fact demonstrates that, similar to adults, children and adolescents with psychiatric disorders are at risk for PNES.

According to Wyllie et al.,<sup>21</sup> in addition to the dissociative disorder, 11 patients (32%) had mood disorders including major depression, bipolar disorder, or dysthymic disorder, usually with severe psychosocial stressors. Eight children (24%) had separation anxiety and school refusal with moderate psychosocial stressors. Two patients (6%) had brief reactive psychosis or schizophreniform disorder.

A few patients each had panic disorder, overanxious disorder, adjustment disorder, oppositional/defiant disorder, or impulse control disorder. Four patients (12%) also had personality disorders. Eleven patients (32%) had a history of sexual abuse. This was especially frequent in the subgroup with mood disorders (7 [64%] of 11 patients).

Although rare, a pure dissociative disorder without other psychiatric diagnosis has already been documented.<sup>11</sup>

Affective and anxiety disorder as primarily etiologic in children with psychogenic seizures,<sup>1</sup> depression and anxiety disorders predominated as primary psychiatric diagnosis, which may explain the generally more favorable response to psychiatric intervention and treatment in our patient group. The goals of treating NESE – to limit the recidivism, to avoid iatrogenic complications, and to ameliorate underlying psychiatric conditions – are best achieved by aggressive diagnosis, orderly initiation of appropriate therapy, and improvement of familial support mechanisms.<sup>26</sup>

## PNES SEMIOLOGY

There is scarce data on seizure semiology in children. However, clinical characteristics of PNES appear to be, in general, similar in children and adults.

In the study of Lancman et al.<sup>8</sup> with 43 children, median seizure frequency was one seizure every 5 days. Clinical characteristics of the seizures varied. However, unresponsiveness with generalized violent and uncoordinated movements involving the whole body (n = 19) or with generalized trembling (n = 11) were the most common features.<sup>8</sup>

In a study of pediatric patients, Holmes et al.<sup>5</sup> categorized PNES according to the type of epileptic seizures that they resemble. These authors found that PNES more frequently resemble generalized tonic-clonic, generalized tonic or partial seizures with complex symptoms. Absence-like or atonic-like are rare and several patients had multiple seizure types, which have been corroborated by others.

Non-epileptic seizures often are distinguished visually by the prolonged nature of the episodes and atypical clinical features.<sup>24</sup> Psychogenic events can mimic genuine status epilepticus and be treated with IV medication and intubation.<sup>20</sup>

## NONEPILEPTIC STATUS EPILEPTICUS IN CHILDREN

Pakalnis et al.<sup>26</sup> demonstrated the importance of a correct diagnosis of this condition to institution of proper therapeutic measures and avoidance of iatrogenic disorders. These authors identified 6 children, among a total of 29 admissions for convulsive status epilepticus in a period of 6 months, who were treated according to an

emergency protocol and whose final diagnosis was psychogenic *status epilepticus*.

Studies in adults suggest that NESE may be a common manifestation of psychogenic seizures.<sup>27</sup> Misdiagnosis can be associated with severe iatrogenic complications and delay of appropriate psychotherapeutic intervention. Children with psychogenic seizures presenting with NESE may be more likely to experience potential iatrogenic problems given that psychogenic seizures are less common in children and are a lower differential diagnostic consideration.<sup>1</sup> Some patients are prone to recidivism of their events until appropriate diagnosis of nonepileptic nature of episodes was made.<sup>26</sup>

### CHILDREN WITH EPILEPSY AND PSYCHOGENIC NONEPILEPTIC SEIZURES

Studies show that 5% to 20% of epilepsy patients have psychogenic seizures.<sup>26,28</sup>

Children and adolescents with epilepsy who present PNES seem to represent a special population with particular characteristics.

In Vincentiis's et al. series,<sup>11</sup> there was no gender predominance and most patients with psychogenic seizures mimicking epileptic seizures were in the first decade of life and first years of adolescence. However, three of their patients under 6 years of age had true psychogenic seizures. It may suggest that children with epilepsy, although not within the expected age range for the occurrence of PNES, deserve special attention, as they experience intense contact with their own condition, as well as that of other patients with epilepsy, because of referrals to epilepsy centers. Therefore, previous history of epilepsy, mostly as a personal history rather than family history, may represent the most important risk factor for PNES in this age group.

By the same token, a special characteristic of this group was the high frequency of patients that mimicked their own epileptic seizures, or some of them.

The frequency of seizures, as well as refractoriness, was not a determinant factor in the occurrence of PNES. In our series, there was predominance of symptomatic/cryptogenic cases and, among these, a predominance of temporal lobe epilepsy. It must be considered that patients with temporal lobe epilepsy have a greater tendency to have a psychiatric disorder than the general population and patients with other types of epilepsy.<sup>29</sup>

In addition, these children may have abnormal EEGs and neuroimaging studies. Based on our experience, in these cases video-EEG is mandatory not only for the diagnosis of PNES but also to elucidate the frequency and differences between epileptic and nonepileptic events. This will help the family and caretakers in distinguishing these events during follow-up avoiding iatrogenic measures.

It also must be taken into account those children with chronic diseases, especially with epilepsy, who experience intense contact with their own condition, as well as that of patients with epilepsy, because of referrals to epilepsy centers.

### CONCLUSION

The importance of diagnosis of PNES in childhood and adolescence is even more significant when one considers that the prognosis in this group is much better than that in adults, probably because of the intervention at an earlier stage of life. This emphasizes the urgency for correct diagnosis, to initiate appropriate therapeutic measures and avoid iatrogenic factors.

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