CASE REPORT

Psychogenic Nonepileptic Seizures in Adolescence: Case Report

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Abstract

Psychogenic non-epileptic seizures are a functional neurological disorder characterized by transient alterations in sensory-motor control and a lack of response to external stimulus, resembling epileptic seizures, but with distinctive semiologic features supported by no ictal activity on electroencephalogram. Since psychogenic non-epileptic seizures can mimic epileptic seizures, children and adolescents with this presentation are usually seen by neurologists. Early diagnosis and intervention with a multidisciplinary approach, including neurology and pedopsychiatry, are critical for limiting unnecessary medical investigations and needless antiepileptic drugs, ascertaining psychiatric comorbidities, and improving treatment adherence and prognosis.

We report the case of a 12-year-old female with several admissions to a neuropediatric inpatient unit with a diagnosis of psychogenic non-epileptic seizures to discuss neurologic and psychiatric assessments, delivery of diagnoses, and planning of outpatient management of this disorder in adolescents.

Keywords: Adolescent; Seizures/diagnosis; Seizures/etiology; Somatoform Disorders/diagnosis; Somatoform Disorders/psychology; Psychophysiological Disorders/diagnosis; Psychophysiological Disorders/psychology

Introduction

A functional neurological disorder is a condition affecting voluntary motor and sensory functions that suggests a neurological disorder but has features incompatible with neurological diagnoses after clinical investigation and thorough neurological examination. Usually, patients with functional neurological disorder experience a significant functional impairment.[1,2] Psychogenic non-epileptic seizures are a subtype of functional neurological disorder whose presentation resembles epileptic seizures, with transient alterations in sensory-motor control and lack of response to external stimulus, but without ictal activity on electroencephalogram (EEG) during episodes.[3,4] Psychogenic non-epileptic seizures can be interpreted as a disturbed neurophysiological response to a threat, such as emotional distress or physiological stressors.[5]

Since most studies have focused on adults,[6,7] there is limited data on the prevalence of functional neurological disorder and psychogenic non-epileptic seizures in pediatric patients. In one study, functional neurological disorder has been estimated to affect 2-4 children per 100,000.[8] Another study considered the prevalence of pediatric psychogenic non-epileptic seizures to be 2-33 per 100,000 and pointed out that its prevalence in children and adolescents undergoing video EEG monitoring was 3.5%-20%.[9] Furthermore, studies in children suggest that a functional neurological disorder diagnosis is most commonly made between 10 and 14 years of age in females and often with multiple and inconsistent symptoms.[10]

Early recognition of psychogenic non-epileptic seizures is crucial to implement an adequate intervention and avoid unnecessary medical procedures or treatment with antiepileptic drugs.[9] Children with frequent episodes can undergo significant academic and social difficulties and cognitive and psychiatric side effects from unnecessary medication.[4] Despite the importance of early detection, the diagnostic delay of psychogenic non-epileptic seizures can take up to 3.5 years, due to multiple factors. As psychogenic non-epileptic seizures generally present as episodes similar to epileptic seizures, patients can be misdiagnosed with epilepsy. Moreover, a lack of resources and access to video EEG, or the reluctance of the family and patient to accept the psychological cause of symptoms, also contribute to delaying a psychogenic non-epileptic seizures diagnosis.[4] Before the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), a traditional approach required

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the exclusion of other neurological or medical causes before considering psychogenic non-epileptic seizures as a diagnosis. This manual adopts a different perspective, including it as a differential diagnosis from the beginning, when seizures have atypical features, persist in spite of adequate treatment with antiepileptic drugs, episodes that are worsened by stress or external factors, and EEG are repeatedly normal, despite the existence of precipitating stressors or the exclusion of feigning.[6,9] This allows for early diagnosis and multidisciplinary interventions, with child neurologists and pedopsychiatrists focusing on the underlying psychopathology that may worsen the psychogenic non-epileptic seizures prognosis.[4,7]

In this article, we report the case of a young adolescent suffering from psychogenic non-epileptic seizures who was repeatedly admitted to a neuropediatric inpatient unit in order to discuss the neurologic and psychiatric assessments, delivery of diagnosis, and planning of the outpatient management of psychogenic non-epileptic seizures in adolescents.

Case Report

A 12-year-old female was admitted to the pediatric emergency department after an acute episode of anomalous oscillatory head movements followed by dizziness, generalized hypotonia, and subsequent fall. The episode was preceded by a sudden loss of vision, a recovery period with somnolence, and the patient reported remembering the event. She was previously healthy, with no known allergies or drug use. Two months before admission, her 5-year-old brother had been diagnosed with severe congenital liver disease with subsequent multiple hospitalizations and was, currently, waiting for a hepatic transplant. Coincidentally, her family noticed behavioral changes, like irritability and aggressive actions toward family members, sadness, hypersomnia, distractibility, decreased school performance, and frequent conflicts with peers and teachers.

During observation in the pediatric emergency department, the patient presented several paroxysmal episodes of a lack of response to external stimulus, with crying, illogical speech, non-focused attention, and rhythmic repetitive axial movements, with pelvic and scapular jerks. Her eyes remained closed during episodes and resisted opening. Each episode lasted 10-15 minutes and responded to intravenous diazepam. Intercritical neurological examinations were normal. At the initial workup, the laboratory data showed normal results, including a complete blood count, ionogram, renal and liver function, lactate, ammonia, C-reactive protein, and negative urine drug screen. Electrocardiogram and cranial computed tomography showed no alterations. The EEG report showed normal background activity and small rhythmic spikes lasting about 15-35 seconds over the left temporal region without clinical manifestations (the patient was right-handed). The diagnosis of inaugural focal epilepsy was made, and she was admitted to the neuropediatric inpatient unit for etiological investigation and treatment. A cranial magnetic resonance imaging was performed and showed no major abnormalities. A lumbar puncture was performed, interpreted as traumatic, showing high protein levels and a normal leukocyte count after sample centrifugation. She manifested similar episodes to those that justified her admission, despite being under escalating antiepileptic treatment. Two subsequent EEG were performed, and its reports displayed persistent abnormal activity, with 4-5 Hz rhythmic spikes, lasting 30-100 seconds over the left temporal region, despite treatment with phenytoin. Given that the inaugural epilepsy coincided with behavioral changes and decreased school performance, the hypothesis of autoimmune encephalitis was considered, and an intravenous immunoglobulin treatment was administered with the reduction of electrical discharges. A second lumbar puncture, again traumatic, showed an elevated cell count and immunoglobulin (Ig) M antibodies for *Borrelia burgdoferii*. Serum antibodies were not detected. For the hypothesis of neuroborreliosis, a 21-day treatment with ceftriaxone was initiated. The patient was submitted to a 24-hour continuous EEG (with clinical monitoring and without video recording, due to the unavailability of the necessary equipment) that reported paroxysmal monomorphic theta activity over the left posterior quadrant, without temporo-spatial evolution, corresponding to a benign electroencephalographic pattern called subclinical rhythmic electroencephalographic discharges in adults. During hospitalization, she had several paroxysmal episodes of tremor in the lower limbs, always in the context of anxiety, and she expressed worries about her brother. An evaluation by a pedopsychiatrist was requested and, although both the patient and her family showed reluctance in accepting a possible psychogenic origin of the symptoms, she was referred to a pedopsychiatry consultation after discharge. Being under antiepileptic drugs, she was also referred to a pediatric neurology appointment. Twelve days later, she underwent another EEG, which was reported to be normal.

One month later, she was readmitted to the pediatric emergency department after an acute episode of rhythmic clonic asymmetrical and non-stereotyped movements of the right upper and lower limbs, sweating, hyperpnea, describing paresthesia of the upper lip and the sensation of 'being outside her own body' (depersonalization). The symptoms began after an argument with someone at school and she had no memory of the episode. She recovered one hour later with somnolence. During observation in the pediatric emergency department, she was calm, with no speech alterations and no meningeal signs. She was admitted to the pediatric ward, where she was evaluated by neurology, infectiology, and pedopsychiatry. Anxiety and depression symptoms were assessed and treatment with gabapentin was started, two antiepileptic drugs were withdrawn, and a new antiepileptic drug was tried, since the report of the 24-hour continuous EEG was still unknown. After a third lumbar puncture and repeating the serologic tests, the hypothesis of an infection was discarded. She was discharged awaiting the results of the N-methyl-D-aspartate receptor (anti-NMDAR) antibodies.

Three weeks later, she was readmitted to the pediatric emergency department after another acute episode of anomalous movements of the upper body, accompanied by hyperventilation and followed by a period of two hours of absence of response. The episode occurred at school after a classmate criticized her and she had no memory of what happened afterwards. She reported 'feeling weird' the previous day, as if 'being outside her own body', and experiencing increasing sadness, more intense since the first hospitalization. Cranial computed tomography and laboratory data including a urine drug screen were performed and were reported normal. She was admitted to the pediatric ward with the diagnostic hypothesis of a psychogenic episode *versus* postictal state.

The hypothesis of autoimmune encephalitis was excluded since the antibodies for anti-NMDAR on plasma and liquor were negative. All previous EEG (including 24-hour continuous) were reviewed and the initially described abnormal activity was reinterpreted as possible normal variations. A diagnosis of psychogenic non-epileptic seizures was assumed and delivered to her and her family by both the pediatric neurologist and pedopsychiatrist, including a thorough explanation of the disorder and clarification of any doubts. The antiepileptic drugs were discontinued, and she started treatment with a selective serotonin reuptake inhibitor, due to a comorbid diagnosis of depressive disorder. She was referred to neurology and pedopsychiatry consultations and the school received information on how to manage future episodes. She remains stable with no subsequent paroxysmal episodes six months after discharge.

Discussion

This report highlights the importance and complexity of a multidisciplinary approach when psychogenic non-epileptic seizures is suspected, emphasizing early detection of clinical findings that allow for its precocious consideration.

In this case, the following signs reliably favored psychogenic non-epileptic seizures over epileptic seizures:

- Episodes preceded by warning signs such as dizziness, blurry vision, sweatiness, and fast breathing;
- Inconsistency of symptoms over time;
- Long duration of episodes;
- Fluctuating course;
- Occasional preserved ictal awareness;
- Presence of external stressors before events and lack of response to antiepileptic drugs.

However, the EEG showed alterations compatible with an inaugural epilepsy, which justified the conducted etiological investigation and antiepileptic drugs treatment. Importantly, EEG can report benign variants, such as subclinical rhythmic electroencephalographic discharges of adults, which are not related to epilepsy but have an epileptiform morphology and can, therefore, be easily misinterpreted. These have a prevalence of 2%-3%, so having different and experienced neurophysiologists interpreting and discussing exams could avoid misdiagnosis and unnecessary treatment, especially when episodes present clinical features that are compatible with psychogenic non-epileptic seizures.[11] A psychogenic non-epileptic seizure diagnosis should rely on careful anamnesis and the observation of episodes, supported by no ictal activity on electroencephalogram during the episodes. In this case, video EEG, which is the gold standard for diagnosis support, could not be performed due to the unavailability of the necessary equipment. Nevertheless, the 24 hours continuous EEG was important in reinterpreting the results of the first exams and contributed to reinforcing the hypothesis of psychogenic non-epileptic seizures. Since psychogenic non-epileptic seizure patients can be sensitive to suggestion, provocation techniques, such as hyperventilation and photic stimulation, can be used to elicit episodes and help in reaching a diagnosis. However, this raises several ethical concerns and, in the

absence of a clear consensus, must be considered on a case-by-case basis. Moreover, the risk of jeopardizing the doctor-patient therapeutic relationship is of particular importance in cases with psychopathology. Therefore, no such techniques were used on our patient.

The ambiguity of the case led to requesting a collaboration with pedopsychiatry during the first hospital admission, with possible precipitating stressors being discussed with the patient and family at an early stage of the investigation. This joint assessment helps in developing a comprehensive neurobiopsychosocial conceptualization of the case, which allows for the design of an individualized treatment plan.[7] Psychological symptoms or psychiatric comorbidities are reported by many patients with psychogenic non-epileptic seizures, and treatment with anxiolytics or antidepressants can be useful when they have a significant impact.[1,8] Our patient suffered from multiple psychiatric comorbidities, namely, anxiety, depression, and dissociative symptoms. Therefore, it was essential to approach and treat these, as they could potentiate maladaptive coping behaviors and negative illness beliefs, which may contribute to worsen symptom severity and treatment response.[1]

Both the patient and family were, from the beginning, reluctant in accepting a possible psychogenic origin of symptoms. This common reluctance emphasizes the importance of having both the pediatric neurologist and pedopsychiatrist communicating cohesively and openly approaching their findings and conclusions, in the key step that is the delivery of the diagnosis. The effectiveness of this communication between the medical team and patient and family has a high impact on the success of the treatment plan.[6,7] Foremost, it is important to name the condition and explain that it is a common and treatable disorder. Relying on metaphors, such as psychogenic non-epileptic seizures being a *'software not a hardware problem'*, can help the patient to better understand the disorder.[12] The patient and family need to acknowledge that the doctor does believe them and does not think they are faking their symptoms. It is also important to give them time to ask questions and address their concerns.[6,12] All of these contribute to improving the patient's confidence in the diagnosis, which is crucial to enhancing their engagement in outpatient management.[7] In the reported case, even though a combined approach with neurology and psychiatry was performed from the beginning, the misleading results of the initial investigation, possibly allied with the reluctance of the family to consider a psychological cause for the symptoms, still led to multiple admissions and exams that should ideally be avoided when dealing with such cases.

In conclusion, psychogenic non-epileptic seizures are a condition in adolescence that spans across neurology and psychiatry. It is associated with significant morbidity and risk of unnecessary medical procedures and pharmacological treatments. Identifying positive examination criteria to reach a rule-in diagnosis within a multidisciplinary medical team is crucial to make the diagnosis, deliver it to the patient and family, and plan outpatient treatment, improving treatment adherence and disease prognosis.

WHAT THIS CASE REPORT ADDS

- Psychogenic non-epileptic seizures is a prevalent condition in childhood that can cause significant morbidity.
- Since psychogenic non-epileptic seizures can mimic epileptic seizures, children and adolescents with this presentation are usually seen by neurologists.
- Psychogenic non-epileptic seizures would benefit from a rule-in diagnosis approach, thereby limiting unnecessary medical investigations and needless antiepileptic drugs.
- Early intervention with a multidisciplinary team (neurology and pedopsychiatry) is critical to improving prognosis and treatment adherence.
- Psychiatric comorbidities must be approached and treated since they can be associated with symptom severity and a poorer outcome.

Conflicts of Interest

The authors declare that there were no conflicts of interest in conducting this work.

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The authors declare that they have followed the protocols of their work center on the publication of patient data.

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Convulsões Psicogénicas Não Epiléticas na Adolescência: Caso Clínico

Resumo:
As crises psicogénicas não epiléticas são uma perturbação neurológica funcional, caracterizada por alterações transitórias do controlo sensório-motor e ausência de resposta a estímulos externos. Assemelham-se, na sua apresentação, a crises epiléticas, tendo características semiológicas distintas e o eletroencefalograma não demonstra atividade ictal durante a sua ocorrência. Uma vez que as crises psicogénicas não epiléticas mimetizam crises epiléticas, as crianças e adolescentes que as apresentam são frequentemente avaliadas por neurologistas. O diagnóstico e intervenção precoces desta perturbação, com uma abordagem multidisciplinar, que inclua a neurologia e a pedopsiquiatria, são essenciais para limitar investigações médicas desnecessárias, evitar a utilização de fármacos antiepiléticos e permitir a abordagem das comorbilidades psiquiátricas, com aumento da adesão ao tratamento e melhoria do prognóstico. Reportamos o caso clínico de uma jovem com 12 anos de idade, com múltiplos internamentos na unidade de neuropediatria, com o diagnóstico de crises psicogénicas não epiléticas. Este caso servirá como plataforma de discussão sobre as abordagens neurológica e psiquiátrica, devolução do diagnóstico e planeamento da intervenção em ambulatório.

Palavras-Chave: Adolescente; Convulsões/diagnóstico; Convulsões/etiologia; Transtornos Psicofisiológicos/diagnóstico; Transtornos Psicofisiológicos/psicologia; Transtornos Somatoformes/diagnóstico; Transtornos Somatoformes/psicologia