INTRODUCTION

Psychogenic Non-Epileptic Seizures (PNES) in children are abrupt onset alterations in behavior that mimic seizures and are often misdiagnosed as seizures even with no abnormal associated epileptiform discharges on electroencephalogram. There have been cases where the true diagnosis of PNES was delayed by several months and the patients were even started on antiepileptic medications. The intractable nature of these episodes and the poor response to treatment prompts the referral to the specialized epilepsy center for inpatient video EEG monitoring unit to capture these episodes. There is paucity of literature about psychogenic non-epileptic spells in children and adolescents resulting in delay of diagnosis, oftentimes by several months. In this review, we discuss the current available literature about the incidence, clinical semiology, diagnosis and management of PNES in this subgroup of patients.

Keywords
- Psychogenic non-epileptic spells
- Conversion disorder
- Epilepsy

Abstract
Psychogenic non-epileptic spells are a form of conversion disorder which is characterized by “seizure like episodes” often misdiagnosed as epilepsy. These are frequently seen in adults with female predominance but occur also in children. The gold standard, for a definitive diagnosis, is inpatient video-EEG monitoring in a specialized epilepsy monitoring unit to capture these episodes. There is paucity of literature about psychogenic non-epileptic spells in children and adolescents resulting in delay of diagnosis, oftentimes by several months. In this review, we discuss the current available literature about the incidence, clinical semiology, diagnosis and management of PNES in this subgroup of patients.

Psychosocial and Clinical profile, Psychiatric features and Predisposing risk factors of children presenting with pseudoseizures.

Bhatia and Sapra conducted a retrospective study in order to determine the psychosocial and clinical profile of children presenting with pseudoseizures. The study was conducted from January 2001 to December 2003 at the University College of Medical Sciences in Delhi, India, and included 50 consecutively admitted children diagnosed with pseudoseizures out of the 110 children (45.5%) with conversion disorders during the study period. Patients received physical and mental status examinations by a psychiatrist and a psychologist, labs, and occasionally video-EEG, Computed Tomography (CT), or Magnetic Resonance Imaging (MRI) as needed. Exclusion criteria included unreliable history, history of epilepsy, febrile convulsions, head injury, mental retardation, or chronic physical illness. Twenty-eight patients were girls (56.0%) with an average age of 9.4 years (range: 7.8-11.6 years), and 22 patients were boys (44.0%) with an average age of 8.2 years (range: 6.8-12 years). Fifty-two percent (N=26) of patients were referred for evaluation of possible pseudoseizures by pediatricians, 24% (N=12) by general physicians, 16% (N=8) from the neurology clinic and 8% (N=4) from the emergency department. The duration from initiation of pseudoseizures to diagnosis was 1-3 months. At presentation, 58% of children (N=29) had received 1 or more anti-epileptic drugs (AEDs) in the past, 24% were still receiving AEDs, 12% were receiving anxiolytic drugs, and 6% were receiving ammonia inhalation.

The most common PNES event semiology was mimicking generalized tonic-clonic seizures. The average event frequency was 5.5 per week and the duration of the events lasted between 10 to 35 minutes. Girls had a longer average duration of symptoms than boys (3.2 months vs. 2.6 months). The most common precipitating factors were social phobia and the fear of examinations (N=15, 30%) followed by quarrels with peers/siblings (N=7, 14%). Sexual abuse occurred in 8% of subjects (N=4). The comorbid psychiatric diagnoses included separation anxiety disorder (N=16, 32%), mood disorders (N=12, 24%), and panic disorder (N=6, 12%), along with other less common diagnoses. All patients were put on drug therapy with anxiolytics and/or antidepressants and/or psychotherapy for 3 months and followed up every 2 weeks for 3 months to assess improvement. At the end of 3 months 36 (72.0%) patients were remitting, 10 (20.0%) were having a decreased frequency of pseudoseizures.
and 4 (8.0%) were not improved. Bhatia and Sapra concluded that children with pseudoseizures had a good outcome with correct diagnosis and treatment [8].

In another study, Patel and colleagues conducted a retrospective review to compare the clinical characteristics of Nonepileptic Seizures (NES) in children younger than 13 years of age to adolescents. All patients with Video-EEG (VEEG) monitoring confirming NES had their medical records and VEEGs reviewed. Exclusion criteria included patients with syncope, breath holding spells, paroxysmal choreoathetosis, and parasomnias. Out of 1,967 patients receiving VEEG monitoring (for a minimum of 23 hours) from April 2002 to December 2005 at the Indiana University Riley Hospital, 3.5% (N=68) had a clinical diagnosis of NES. Typical spells were recorded for 59 of 68 patients and were included in the study.

Twelve years 9 months was the mean age of onset (SD: 3 years 1 month; 5.5 years-19.5 years). At the time of VEEG diagnosis, the mean age was 13 years 4 months (SD = 3 yr 1 month; 5 years 10 months-20 years). There were in 22 patients less than 13 years (group A) and 37 patients 13 years and older (group B). There was a female predominance in group B (2.3:1 with 26 females) and there was no difference in the female to male ratio in group B (1:1 with 11 females). In group A, subtle motor activity, including staring, head shaking, generalized limpness, behavioral changes, eye fluttering, and oromotor activity, was the most common manifestation of NES (p<0.01; N=19). In group B, prominent motor activity, including generalized jerking, focal motor activity, complex motor activity, and generalized tremor, was the most common manifestation of NES (p<0.001; N=33). Frequent stressors in both groups included family discord, school, and interpersonal conflicts. The least frequent stressor was sexual abuse, reported in 4.5% of patients (N=1) in group A and 5.4% (N=2) in group B. There were no significant differences between groups in terms of physical abuse, school difficulties, psychiatric illness, neurologic illness, or headaches. In group A, cognitive dysfunction (p<0.001) and epilepsy (p<0.01; partial epilepsy in 80% of group A and 48% of group B) were more common. In group B, depression was more common but not statistically significant (group A=9%, group B=35%). Patel et al concluded that young children and adolescents at risk for developing NES may be identified by differences in clinical semiology and predisposing factors [5].

Metrick et al conducted a retrospective chart review study of children less than 16 years of age admitted for intractable epilepsy evaluation. From August 1986 to August 1998, 27 of 220 children (12%) were evaluated and were subsequently found to have nonepileptic events by video EEG monitoring (lasting ≥ 24 hours) at the MINCEP Epilepsy Program at Gillette Children's Hospital in St. Paul, Minnesota. There were 18 females and 9 males. Patients had a median age of 8.4 years (7 months-16 years). The nonepileptic events were categorized into four groups including pure psychogenic events (N=5), psychogenic events plus epileptic seizures (N=3), pure nonepileptic physiologic events (N=5), and nonepileptic physiologic events plus seizures (N=14).

Unusual reactions to environmental stimuli, such as staring spells, repetitive stereotypic behaviors, and abnormal movements secondary to abnormalities in muscle tone, were misinterpreted by parents as seizures and were the most frequent nonepileptic physiologic events. Patients had a history of multiple seizure types in all groups except for those with pure psychogenic seizures. These were later defined as paroxysmal choreoathetosis in two patients, one of which was attributed to medication in one patient. Sixty-four percent of patients (N=14) had a history of status epileptics. This included 11 of 14 patients with physiologic events plus seizures. Children with nonepileptic physiologic events were more likely to have abnormal findings on neurologic examination. Twenty-five of 27 total children (93%) had a history of interictal epileptiform events on previous routine EEGs. AEDs were discontinued in 8 patients (30%) after confirmation of nonepileptic events, most commonly in the physiologic group (4/5) and least commonly in the psychogenic + seizures group (0/3). Nine patients (33%) had the numbers of AEDs reduced, with 8 of these belonging to the physiologic + seizures group (8/14). Metrick et al concluded that intensive video EEG monitoring is the preferable method for identifying nonepileptic events due to the diagnosis being confounded by abnormal findings on routine EEG, neurologic examination, and neuroradiologic studies [9].

Watemberg et al reported on the usefulness of adding video recording to routine EEG studies in infants and children with frequent paroxysmal events. Over a 4-year period, video recording was added to a routine EEG in 137 of 666 cases. Video was added to EEG analysis for paroxysmal eye movements, tremor, suspected seizures, myoclonus, staring episodes, suspected stereotypias and tics, absence epilepsy follow-up, cyanotic episodes, and suspected psychogenic nonepileptic events. Video was added to EEG in 7 infants 0-2 months of age, 19 infants 2-12 months of age, 33 toddlers 1-3 years of age, 48 preschool and school-age children 4-10 years of age, and 30 children older than 10 years. The mean patient age was 4.8 years (range: 2 days-18 years). The nature of the event was determined in 45% (N=61) of video EEGs referred for frequent paroxysmal events. The highest proportion of helpful contributory studies was in the 1-3 year old group (61%, N=20) and was least helpful in the 0-2 month olds (0%). Hospitalized patients made up 28%. Video recording was not helpful in 60 cases due to the patient not experiencing a clinical event. The average duration of the video EEG was 2.6 minutes (SD: 12.7 min) with 20 cases of stereotypias and paroxysmal eye movements solving the clinical question in <20 minutes. Video recording helped confirm or rule out epilepsy in 18 of 78 studies with the indication ‘rule out seizures’. Nine patients were referred for new, different spells with known concomitant epilepsy and all 9 were shown to be epileptic seizures. The other 9 cases were comprised of 3 children diagnosed with epilepsy on video-EEG and 6 cases of non-epileptic events. On average it took 21.7 minutes (SD: 6.4 min) to confirm or rule out seizures when there was additional information in the patient record and 26.6 minutes (SD: 5.7 min) when patient records did not have any information to help confirm or rule out seizures. Video EEG was particularly useful in psychogenic nonepileptic events, paroxysmal eye movements, staring spells, myoclonic jerks, and stereotypias. Watemberg et al concluded that the addition of video recording to routine EEG is effective, cheaper, less time consuming, and more comfortable to the patient and his or her family [10].

Wyllie and colleagues conducted a retrospective study...
of the psychiatric features of children and adolescents with pseudoseizures. From 1992-1996 at the Cleveland Clinic Foundation, 38 children and adolescents were identified with pseudoseizures diagnosed by ictal video EEG. Inclusion criteria required that subject be evaluated by a child psychiatrist, causing 4 patients to be ruled out. The 34 patients included in the study had a mean and median age of 14 years (range: 9-18 years). The mean and median age of onset of pseudoseizures was 13 years (range: 8-18 years). There was a median of 4.5 months (mean: 11.4 months; range: 0.5-48 months) of the duration of pseudoseizures until video EEG diagnosis. Twenty-one patients had daily pseudoseizures, 10 patients had weekly spells, and 3 patients had spells less frequently.

At the time of diagnosis, 70% of patients (N=24) were taking AEDs. A diagnosis of mood disorders including major depression (N=6), bipolar disorder (N=1), and dysthymic disorder (N=4) occurred in 32% of patients (N=11: 9 females, 2 males). Eight of 11 patients with mood disorders also had severe psychosocial stressors. Twenty-four percent (N=8: 6 females, 2 males) had moderate psychosocial stressors and separation anxiety issues. Brief reactive psychosis or schizophreniaform disorder occurred in 6% of patients (N=1 each). Twelve percent (N=4) had personality disorders, most commonly dependent traits and borderline personality disorder with 2 patients each. A diagnosis of panic disorder, overanxious disorder, adjustment disorder, oppositional/defiant disorder, or impulse control disorder occurred in 1-3 patients each. A history of sexual abuse was present in 32% of patients (N=11), most frequently in the subgroup with mood disorders (7 of 11 patients [64%]). A history of physical abuse was present in 6% of patients (N=2). Fifteen patients (44%) had severe family stressors including death of a close family member, parental discord, or recent parental divorce. Axis III disorders included epilepsy (N=4), single febrile seizures (N=1), severe head trauma (N=1), and hearing impairment (N=1). Twenty-one patients were reached for telephone follow-up and 15 of those (72%) were free of pseudoseizures from the preceding 9-55 months (mean: 30 months). Wyllie et al concluded that in children with pseudoseizures, major mood disorders and severe environmental stress, especially due to sexual abuse, were common and should be considered. Video-EEG and detailed pediatric psychiatric assessment are critical in the diagnosis and treatment of pseudoseizures [11]. Vincentiis et al conducted a prospective observational study to evaluate the outcomes after psychogenic seizures after diagnosis. These studies showed that only 25 to 38 percent of patients achieve full recovery from this condition [13-17]. However, outcomes have been reported much more favorable in children [18-20].

Wyllie et al conducted a prospective observational study to determine the outcomes after psychogenic seizures were documented by video-EEG monitoring. From 1980 to 1988 at the Cleveland Clinic Foundation there was a total of 21 nonepileptic children who had psychogenic seizures during video-EEG recording. Video-EEG monitoring lasted for at least 8 hours in each patient. Children with nonepileptic paroxysmal behaviors, including parasomnias, hyperventilation attacks, breath-holding spells, syncope, migrainous disorders, movement disorders, and gastroenterologic phenomena, were excluded. The ages ranged from 8-18 years of age (mean: 14.5 years), with eight 10-13 years olds and thirteen 14-18 year olds. Seventy-one percent (N=15) were female while 29% were male (N=6). The mean onset of episodes was at 13.8 years (range: 8-18 years) with a mean time from onset to diagnosis of 7 months (range: 1 week-5 years). Patients had an average episode frequency from 1-8 per day. Seventy-six percent of patients (N=16) were taking AEDs. Eight patients had been previously hospitalized for seizure control, 5 had drug side effects, and 2 had undergone invasive diagnostic procedures. Several patients and their families had previously been told that the episodes may be psychogenic and received psychological counseling that was unsuccessful in affecting the episode frequency. Frequency precipitating psychosocial stress factors included living with significant family discord (N=9), A history of mood disorders was the most common psychiatric diagnosis (N=13, 61.9%), followed by pure dissociative disorder (N=3, 14.3%), conduct disorder (N=2, 9.5%), pure oppositional-defiant disorder (N=2, 9.5%), and hyperkinetic disorder (N=1, 4.8%). Epilepsy was current in 19 patients (90.5%), and 9 were cryptogenic. 8 were symptomatic (N=7 mesial temporal sclerosis, N=1 frontal lobe lesion), and 2 were idiopathic. Epilepsy was well-controlled in 2 patients (9.5%). Seizure frequency was daily in 36.8% of patients (N=7), weekly in 26.3% (N=5), and monthly in 42.1% (N=8). Sixteen of 21 patients were on AED treatment, with 8 patients (42.1%) on one AED, 6 (31.6%) were on two AEDs, and 3 (15.8%) were on 3 AEDs. A family history of epilepsy was present in 9 (42.9%) patients. There was a history of sexual or physical abuse in 19% (N=4) of patients and a history of psychological abuse in 3 patients (14.3%). Eleven (52.4%) of the patients were noted to have an inadequate family setting contributing to a stressful home environment. PNES semiology was most commonly events that mimicked their own epileptic seizures (47.6%, N=10), dissociative episodes that did not resemble their epileptic seizures (42.8%, N=9), and panic attacks (9.5%, N=2). After the diagnosis of PNES and referral for psychiatric treatment, 10 patients (47.6%) started psychoactive drug therapy. 3 patients (14.4%) had their previous medications adjusted, and the number of AEDs were reduced in 6 patients (28.6%) [12].

**Outcome of PNES in Children and Adolescents**

There were several studies that assessed the prognosis of psychogenic seizures after diagnosis. These studies showed that only 25 to 38 percent of patients achieve full recovery from this condition [13-17]. However, outcomes have been reported much more favorable in children [18-20].

Wyllie et al concluded that in children and adolescents with pseudoseizures during video-EEG recording, Video-EEG monitoring lasted for at least 8 hours in each patient. Children with nonepileptic paroxysmal behaviors, including parasomnias, hyperventilation attacks, breath-holding spells, syncope, migrainous disorders, movement disorders, and gastroenterologic phenomena, were excluded. The ages ranged from 8-18 years of age (mean: 14.5 years), with eight 10-13 years olds and thirteen 14-18 year olds. Seventy-one percent (N=15) were female while 29% were male (N=6). The mean onset of episodes was at 13.8 years (range: 8-18 years) with a mean time from onset to diagnosis of 7 months (range: 1 week-5 years). Patients had an average episode frequency from 1-8 per day. Seventy-six percent of patients (N=16) were taking AEDs. Eight patients had been previously hospitalized for seizure control, 5 had drug side effects, and 2 had undergone invasive diagnostic procedures. Several patients and their families had previously been told that the episodes may be psychogenic and received psychological counseling that was unsuccessful in affecting the episode frequency. Frequency precipitating psychosocial stress factors included living with significant family discord (N=9),
having a recent family death (N=6), having an alcoholic parent (N=4), having a parent with significant medical illness (N=2), having major problems with peer relations (N=2), or having been sexually assaulted (N=1).

During video-EEG monitoring, patients had a mean of 2.2 recorded seizures (range: 1-8). Seizures occurred spontaneously in 15 patients (71%) and in response to suggestion and intravenous saline injection in 6 patients (29%). Patients or family reported that all videotaped episodes were the same as their habitual episodes. Episodes lasted 1-17 minutes (mean: 2.4). Episode semiology in 10 patients (48%) included 20 episodes of unresponsiveness, generalized limb jerking, and thrashing movements. All patients were amnesic after episodes. Three patients reported auras prior to episodes, including numbness, tingling, or light-headedness. After diagnosis of psychogenic seizures, 13 patients had outpatient treatment (mean: 9 months; range 3-36 months), 8 patients had inpatient treatment (mean: 2 months; range 0.5-13), 5 patients had both, and 1 patient refused to discontinue AEDs. Psychotropic medications (imipramine, desipramine, or haloperidol) were used to treat 5 patients. Eighteen patients were reached during telephone follow-up up to 66 months after diagnosis (mean: 30). Twenty-two percent (N=4) had ongoing episodes and 78% (N=14) were episode free. Fifty-seven percent (N=8) had no episodes after diagnosis and 43% of patients had their episodes decrease gradually over a span of 3 to 48 months prior to coming episode free [18].

In another study by Wyllie and colleagues, a retrospective study was conducted to compare the outcomes of psychogenic seizures in children and adolescents to adults. Subjects were identified through video-EEG report review. Twenty pediatric patients 18 years old or younger were identified and two were excluded because they couldn’t be reached for follow-up. Fifty-three adults age 25 or older were identified and 20 were included. Thirty-three patients were excluded. Twenty patients were not able to be located for following up, 9 patients had a history of self-mutilation/psychosis/physical violence/current litigation with the hospital, 3 patients declined to be involved in the study, and 1 patient was excluded due to death. Structured telephone interviews were conducted to collect follow-up data at 6 months, 1 year, 2 years, and 3 years after diagnosis.

The median age in children and adolescents was 14.5 years (range: 8-18) and 34.0 years in adults (range: 25-56). It was rare that children under 10 years old had psychogenic seizures, and during psychogenic seizures were more likely to have mannerisms, parasomnias, hyperventilation attacks, breath-holding spells, syncope, or movement disorders. Sixty-seven percent of the younger group were female compared to 85% in the older group, and the younger group was 33% male versus 15% male in the older group (p=0.173). The age at onset of psychogenic seizures was a median of 14.0 years in the younger group and 30.5 years in adults. The frequency before diagnosis was greater than 1 “seizure” per week in 89% (N=16) in the younger group and 60% (N=12) in the older group. After the diagnosis of psychogenic seizures, the events resolved completely in 44% (N=8) of the younger group and 20% (N=4) of the older group (not significant). Younger patients had significantly better outcomes in terms of the percentage seizure-free compared to adults at 1 year (73% vs. 25%; p=0.106), 2 years (75% vs. 25%; p=0.001), and 3 years (81% vs. 40%; p=0.13). After diagnosis, 14 of the younger group (78%) and 10 of the older group (50%) obtained psychological counseling. However, despite recommendations to discontinue AEDs 1 pediatric patient (6%) and 6 adults (30%) continued taking AEDs. The patients that obtained psychological counseling did not have a correlation with outcome. The definitive diagnosis obtained by video-EEG was critical to the resolution of psychogenic seizures in many patients [19].

Gudmundsson et al conducted a retrospective chart review study to report a survival analysis of children and adolescents with pseudoseizures and without concomitant epilepsy. From 1988 to 1994 at Birmingham Children’s Hospital, UK, the records of all children and adolescents with pseudoseizures without epilepsy referred and treated in the psychiatric ward (inpatient or day-patient) were reviewed. During inpatient admission, treatment included milieu therapy and attendance at the school combined with anticonvulsant withdrawal. EEGs were performed in all children. A definition of “cure” was 6 months free of seizures. Seventeen children and adolescents were identified with a mean age at presentation of 12 years 9 months (SD: 26 months; range: 8 years 3 months to 15 years 9 months). There were 2 males and 15 females. The DSM-IV criteria for somatization disorder were not fulfilled in any subject. Fourteen weeks was the mean inpatient stay for 12 children (SD: 9 weeks) and 25 weeks (SD: 17 weeks) for 11 day patients. 6 patients were seen both as inpatients and day-patients. The duration of symptoms until diagnosis ranged from 1 to 59 months (mean: 12 months; SD 15 months).

Spell characterizations varied and multiple subjects had more than 1-3 types (median: 2; SD 0.79). The mean spell frequency was 18 spells per week (range: 1-70/wk). The most common spell type was the ‘swoon’ (N=10), followed by ‘trashing’ (N=9). Eight subjects received a diagnosis of epilepsy and were prescribed AEDs on presentation. AEDs were weaned over a period of 6 weeks to 9 months after diagnosis of pseudoseizures (mean: 12 weeks). Three patients were lost to follow-up due to becoming too old for the service or referral to other health care locations. Eighty-two percent of patients (N=14) recovered and resumed regular school attendance. Fifty-nine percent of subjects were spell free at 6 month and 63% were spell free at 1 year. The mean symptom survival time following treatment was 1.5 years. Recovery followed an exponential distribution. Better outcomes were associated with female sex, having more types of spells, not receiving both inpatient and outpatient treatment, and younger age at presentation. Gudmundsson et al concluded that prompt identification may be important in ensuring a good outcome due to the deteriorating prognosis with age at treatment [20].

CONCLUSION

PNES in children is a most challenging condition in terms of both diagnosis and treatment. Once the diagnosis is established, information needs to be conveyed to both the family and the patient in a non-judgmental manner. There may be instances when parents or patient need more information and a longer clinic visit to accept the diagnosis. Patients with psychogenic spells should be referred to a psychiatrist or psychologist to determine appropriate treatment plan which includes...
psychotherapy and possible psychotropic medication to treat co-morbid conditions. [21-26]. Prognosis in children is much better than in adults and a significant percentage of children become symptom free several years after the diagnosis is made [19,20].

REFERENCES