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Original Article

Parental Ranking of Terms Describing Nonepileptic Events

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ABSTRACT

Paroxysmal nonepileptic events of psychogenic etiology in children and adolescents are common. Patients and their parents are often confused by the terminology used to describe these events. This can lead to frustration and may result in the failure to obtain the necessary nonpharmacologic treatment. Various terms are used to describe such events, some of which might be considered offensive to some individuals. Surveys from 146 parents or guardians of patients identified from a general pediatric clinic, a general neurology clinic, and a pediatric epilepsy monitoring unit were completed with the aim of determining which words and phrases were least offensive. It was determined that nonepileptic events, functional seizures, and nonepileptic attack disorder were the least offensive labels; whereas "it is all in his or her head," hysterical seizures, and psychogenic seizures were the most offensive terms. This is the only study of its type in the pediatric population. Although each child and family requires individualized communication, we hope that this article will provide useful information to guide the practicing pediatric neurologist in informing families that their children are having events of nonepileptic etiology.

Introduction

Paroxysmal nonepileptic events of psychogenic etiology (e.g., conversion disorder) have a prevalence estimated at 2 to 33 per 100,000 [1]. These events constitute up to 5-20% of the diagnoses of intractable seizures and referrals to epilepsy centers as well as 10-40% of admissions to epilepsy monitoring units [2-8]. Many reports have specifically addressed these events in the pediatric population [2-4,7-14]. As these events can resemble epileptic seizures, confusion can result when patients and families are informed these episodes are nonepileptic and antiepileptic medications are not needed. Some patients have already received an erroneous diagnosis of epilepsy and are understandably resistant to a psychogenic etiology for their nonepileptic events. This resistance may lead them to seek epilepsy-oriented care, to continue to take antiepileptic drugs, or to refuse to seek psychological evaluation and treatment [4,6,9,10,14,15]. Furthermore, communication as to the correct diagnosis as early as possible in the course of the symptoms is related to more effective therapy. It is well established that those who receive the diagnosis earlier in the course of their symptoms have better outcomes than those diagnosed later [9,16,17]. Children are usually diagnosed much faster than adults, oftentimes in months rather than years, and have a better outcome of event freedom than adults [9,14,18].

Recently as many as 15 labels have been described in the literature to identify nonepileptic events of psychogenic etiology. These include pseudo-seizure, nonepileptic attack disorder, nonepileptic seizure, nonepileptic event, stress-related seizure, functional seizure, psychogenic seizure, psychoseizure, pseudo-epileptic attack, hysterical seizure, psychogenic attack, hysterical attack, pseudo-epilepsy, hysteroepilepsy, and hysterical epilepsy [12,19]. Many authors have put forth their arguments for adopting various terms, yet only one study has been published aimed at identifying patient preference for the labels used

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to describe these events in adult general neurology patients [20].

The aims of this prospective, questionnaire-based study were to determine (1) which labels describing paroxysmal nonepileptic events parents or guardians find the most and least offensive, (2) whether these responses varied by the clinical setting in which the child was seen, and (3) whether parental decision making would change once the diagnosis was amended. With a better understanding of family perception of the labels for paroxysmal nonepileptic events, terminology can be used that might facilitate acceptance of the diagnosis and need for psychological evaluation and treatment. To the best of our knowledge, this is the first study of this type in a pediatric population. To allow for comparison, this study was modeled off the adult study by Stone et al. [20].

Methods

After approval was obtained from the institutional review board, English-speaking patients were identified from a general pediatrics resident clinic, a general neurology outpatient clinic, and a pediatric epilepsy monitoring unit in a community-based, free-standing children's hospital. At the end of the ambulatory visit or during admission to the pediatric epilepsy monitoring unit, parents or guardians reviewed and completed a questionnaire regarding labeling of paroxysmal nonepileptic events. The questionnaire presented two scenarios. Scenario 1 stated "Imagine that your child has episodes of blackouts or shaking. The doctor tells you that all of the tests are normal. Please rank the following terms in the order of phrases that bother you most/are most offensive (number 1) to phrases that bother you least/are least offensive (number 12) if they were given to you by your doctor to explain the diagnosis of your child." These 12 terms are listed in Table 1.

Scenario 2 stated "Now imagine that your child has been having blackouts or shaking. You have been told they are having seizures, and they have been started on medication. You go to see a specialist who performs additional tests. The specialist tells you these tests are normal, that your child is not having *epileptic* seizures, and your child no longer needs medication. Keeping in mind this new situation, again rank the following terms in the order of phrases that bother you most/are most offensive (number 1) to phrases that bother you least/are least offensive (number 12) if they were given to you by your doctor to explain the diagnosis of your child." In addition, following this scenario, parents were asked four questions (Table 2) aimed at evaluating their reactions to the information the specialist provided them in scenario 2.

The averaged rank responses to each label were calculated (Table 3). As shown in Table 3, there is a clear difference in the numeric ratings for each descriptor. The terms were clustered into three groups (most offensive, least offensive, middle-neutral) in order to increase the statistical power of the comparisons. These groups were clustered by means that are statistically different from one another (data not shown).

Table 1. Labels included in the questionnaire for scenarios 1 and 2, which parents were asked to rank from 1-12 in order from most offensive (1) to least offensive (12)

All in his/her head
Nonepileptic events
Tonic-clonic seizures
Functional seizures
Hysterical seizures
Pseudo-seizures
Paroxysmal episodes
Grand mal seizures
Epilepsy
Nonepileptic attack disorder
Stress-related seizures
Psychogenic seizures

Table 2. Patient responses to follow-up questions contained in the questionnaire, asking for parental opinions based on the information contained in scenario 2

Question	Yes (n %)	No (n %)	Don't Know (n %)
Would you believe the doctor?	44 (30%)	45 (31%)	55 (38%)
Would you think the doctor is crazy?	21 (14%)	103 (71%)	21 (14%)
Would you seek another opinion?	121 (83%)	6 (4%)	17 (12%)
Would you continue to give your child medication?	40 (28%)	52 (36%)	53 (36%)

The averages for each of the three groups were compared for scenarios 1 and 2, as well as according to the patient population (general neurology, general pediatrics, and pediatric epilepsy monitoring unit). The terms epilepsy, tonic clonic seizures, and grand mal seizures were excluded from analysis because these each describe epileptic events and would not be used to label events that were demonstrated to be nonepileptic. These terms were initially included in the questionnaire to allow comparison of this pediatric study to the adult study by Stone et al. [20].

In order to test the results for generalizability between the patient populations (general neurology, general pediatrics, and pediatric epilepsy monitoring unit), a one-way analysis of variance with Bonferroni post hoc test was performed (data not shown, available upon request). Descriptive measures (means) were used to evaluate parent participant responses to four questions (Table 2) to aid in assessing what impact a diagnosis of nonepileptic events had on parental behavior.

Results

One hundred seventy-seven questionnaires were distributed and 146 were used for data analysis. Ten questionnaires were not returned, and 21 did not rank every label with 1 through 12 continuously, thus making them invalid for analysis.

Review of the mean responses for each term (Table 3) reveal that the single most offensive term for both scenarios 1 and 2 was it is all in his/her head. This along with the next two most offensive terms (hysterical seizures and psychogenic seizures) (Table 3) were grouped into group 1 for further comparison.

Overall, the least offensive term for all patients in both scenarios was nonepileptic events (Table 3). This term, in addition to nonepileptic attack disorder and functional seizure, constituted group 2.

The remaining terms (stress-related seizures, paroxysmal seizures, pseudo-seizures) were identified as moderately offensive and were combined into group 3.

A comparison performed among groups 1, 2, and 3 for all responses in scenarios 1 and 2 reveals a significant difference between the groups ($P < 0.001$ for scenario 1 and scenario 2, data not shown, available upon request), indicating that the terms in group 2 are statistically significantly the least offensive. Furthermore, the different patient populations were not significantly different from one another with regard to the rankings of the terms (data not shown, available upon request).

Of note, the ranking of labels in scenarios 1 and 2 were not statistically different and furthermore the labels were ranked in the same order with the exception of the terms paroxysmal episodes and stress-related seizures, which switched in rank preference (Table 3). This indicates that

Table 3. Mean responses for each questionnaire term stratified by scenario and patient population

Term	Scenario 1	Scenario 2	Scenario 1			Scenario 2		
	All Patients (n = 146)	All Patients (n = 146)	Neuro (n = 72)	Gen Peds (n = 36)	PEMU (n = 38)	Neuro (n = 72)	Gen Peds (n = 36)	PEMU (n = 38)
Nonepileptic events	**8.2**	**8.0**	**8.5**	7.0	**8.8**	**8.5**	6.4	**8.5**
Nonepileptic attack disorder	7.5	7.4	7.5	7.4	7.6	7.4	**7.5**	7.2
Functional seizures	7.5	7.3	7.4	**7.5**	7.6	7.2	6.9	7.9
Paroxysmal episodes	7.4	6.9	7.4	7.0	7.9	7.0	6.7	7.1
Stress-related seizures	7.0	7.0	7.0	6.6	7.6	7.0	6.3	7.6
Pseudo-seizures	6.4	6.3	6.3	6.5	6.5	6.2	6.4	6.4
Psychogenic seizures	5.7	6.1	5.8	5.6	5.6	6.2	6.4	5.5
Hysterical seizures	4.3	5.0	4.0	5.3	4.0	4.7	5.8	4.6
All in his/her head	1.9	2.1	1.7	2.5	1.7	1.8	2.9	2.0

Abbreviation:
PEMU = Pediatric epilepsy monitoring unit
Note: Least offensive terms, as determined by the highest mean, are bolded for each subset.

parental preference for labels was not altered once the diagnosis was amended.

Parental responses to four follow-up questions were reviewed (Table 2). Twenty-eight percent responded that they would continue giving medication even after being told medications were not needed, while 36% were unsure if they would continue giving medication. In addition, many (83%) would seek a second opinion after being told that the events being experienced by their child were nonepileptic and did not require medication.

Discussion

In the study of adult general neurology clinic patients by Stone et al. [20], it was determined that the terms nonepileptic attack disorder, psychogenic seizure, and pseudoseizure each offended about one third of the patients, whereas the terms functional seizures and stress-related seizures were significantly less offensive ($P < 0.0001$, Fisher's exact test) than the other three terms. The term functional seizures was included in group 2 as least offensive in our study; however, nonepileptic attack disorder was ranked as one of the least offensive labels in our study and is thus in direct opposition with the results found by Stone et al. This illustrates the difficulty in standardizing terminology.

As the terms in group 2 are significantly different from those in groups 1 and 3, consideration should be given to make these the preferred terms when referring to nonepileptic spells of psychogenic origin. Ultimately, however, the manner in which this information is conveyed is best determined by the practitioner based on his or her knowledge of the individual needs of the family. The current study demonstrates that terms can be clearly differentiated into those that are least offensive (group 2) and those that are most offensive (group 1). As the mean rankings of individual terms in each of the three patient populations do not display a statistical significant difference, it can be inferred that these results could be generalized to several pediatric populations.

The expressions psychogenic nonepileptic seizures and psychogenic nonepileptic attacks remain in common usage [21,22]. This is despite the previous data that indicate patients find the terms functional seizures and stress-related seizures least offensive [13]. However, these have not been routinely incorporated into the medical community. Although this terminology now includes the word nonepileptic, the commentary discussing these labels relates continued confusion and frustration of patients who are trying to understand their diagnosis.

The importance of determining a term that is most descriptive with the least negative connotation is not merely semantic as it could have a significant outcome on patient acceptance of the diagnosis. As identified in children, many patients suffering from paroxysmal nonepileptic events have associated psychological issues and require mental health intervention [4,7,13]. There are also case reports of an association with sexual or physical abuse in children [2,7,9,11,13] and adults [5,16,18]. If a patient or family does not have insight into the diagnosis, it ultimately delays appropriate therapy for the patients and leaves them in a potentially dangerous situation. However, terms such as nonepileptic state what an event is not, whereas inclusion of a term that indicates etiology such as psychogenic could lead a family to seek mental health care more rapidly. It should be noted that the diagnosis of nonepileptic events might require testing that includes electrophysiology, neuroimaging, electroencephalogram, and metabolic and psychological evaluations. Proper communication of a nonepileptic event diagnosis may prevent retesting and therefore have a significant cost savings.

Adult patients' understanding and reaction to the diagnosis of nonepileptic events and the impact on outcome was previously investigated [15]. It was found that a third of the patients had some understanding of the diagnosis, but even these patients were confused as to the nature of the events because many lacked a temporal relationship to an identified stressor. However, 21% indicated relief at not having epilepsy. Of the 77% of patients referred for mental health evaluation who saw a psychologist, 24% stated no benefit. In the end, the study concluded that the current level of education and psychological support is inadequate, and that a patient's response to a diagnosis of nonepileptic events may predict outcome and may be helpful in tailoring the therapeutic approach.

However, undergoing psychological counseling does not always correlate with better outcome in pediatric patients [7,13,14], and some studies have indicated that psychotherapy may be more beneficial in adults [23,24]. One of the best predictors of outcome in both children and adults appears to be reinforcement of the diagnosis over time

[13,15]. By using acceptable terminology along with reinforcement of diagnosis, families and patients can feel confident about their diagnosis and seek necessary mental health services. It should be recognized that patient and family preference for terminology might vary with gender, socioeconomic status, current or past neurological diagnoses, and ethnicity. Given recent reports of the gap in including Hispanic individuals in large-scale studies, this population needs to be considered more carefully in the future [25].

Results to the questions aimed at assessing parents' or guardians' reactions when a diagnosis of nonepileptic events was given after an initial delay in diagnosis can be seen in Table 2. Parents indicated a resistance to the diagnosis of a nonepileptic event and cessation of previously started antiepileptic drugs. Only one third of parents indicated that they would accept the diagnosis of a nonepileptic event when they had previously been told their child had seizures. The other two-thirds either frankly disbelieved the new diagnosis or expressed doubt. This could indicate the combination of the power of the initial diagnosis and the effect of parents treating their children, taking into account the known possible adverse effects of antiepileptic drugs. This reticence to accept the diagnosis of a nonepileptic event quickly is most unfortunate because, as previously reported, a shorter duration of nonepileptic events prior to diagnosis is associated with a better outcome and freedom from nonepileptic events in the future [13,17].

It is interesting to note that the majority of the respondents (71%) did not believe that the second physician was incompetent ("crazy"), but most likely incorrect. It appears that the strategy that most families would use to deal with the disparity in diagnosis was a third opinion (83%). However, the scenarios did not provide any information regarding the evidence that was used to make either diagnosis (e.g., video electroencephalogram). The high percentage of responses from parent responders indicating they would not believe the diagnosis or would seek a second opinion suggests that delayed diagnosis may prolong antiepileptic drug usage and acceptance of diagnosis, inferring a poorer outcome for patients from previous studies [14,15]. Because it is not uncommon for physicians to be wary of stopping antiepileptic drugs that have been started in patients with nonepileptic events [6], it is understandable that many parents responded they would be unwilling to stop medication previously prescribed to their child, even when told to discontinue by a specialist (Table 2). This response underscores the importance of a correct, early diagnosis and a forthright discussion about that diagnosis, recognizing that both physical and sexual abuse may be contributing to the nonepileptic events. In addition, other potential age-related factors such as difficulty at school and family dysfunction should be considered. Even if medication is withdrawn, the neurologist should stay involved for purposes of continuity and the possibility of a seizure occurring as a comorbidity with the nonepileptic event.

Thus although the term nonepileptic event was the least offensive term across all groups and both scenarios, this nomenclature is not currently the most preferred in the literature when communicating with patients and families. Whether this term or another is adopted as the preferred descriptor is important, as patients and families should have insight into their diagnosis through the nomenclature used to guide them in getting the proper treatment. Although seemingly logical, it remains to be proven that family reaction to a term should be used as the critical determinant of which term is used. Furthermore, a prospective study would need to be performed to determine if the term, not to mention the manner in which it was described, is a significant predictor of long-term outcome.

It is apparent that there are many more questions raised than answered by the current study that was intended to provide a single piece of information—what terms parents find most and least offensive with regard to nonepileptic events. There are no real data to indicate how the use of terms affects outcomes. It is unknown if a more descriptive, but possibly more offensive term, such as psychogenic, facilitates or retards acceptance of diagnosis and appropriate treatment.

The strengths of this study include its prospective design, standardized questionnaire, and utilization of statistical methods in order to determine which groups of terms clustered with regard to "offensiveness" to the parent. Furthermore, several populations of patients or parents were queried in order to determine if responses varied by the clinical scenario and thus could be generalized to a variety of clinical settings. Finally, this is the only study that has attempted to address the issue of nonepileptic event terminology in the pediatric population.

However, there were several limitations to the present study. Because this was only the second study investigating patient or parent preferences for labels describing nonepileptic events, and the first pediatric study, it was modeled against the one currently in the literature by Stone et al. that studied an adult population. As such, similar labels were included for parents to rank, including labels that clearly describe epileptic events that were then omitted in analysis. In addition, this study was performed in written form, while the participants in the study by Stone et al. had questions about preference read to them. It is possible that this could have an impact on preference, both by hearing labels aloud or by lack of ability for pronunciation or emphasis when reading a written word. Future studies should address the understanding of the respondents for the terms used, taking into account factors such as education level and socioeconomic status.

The findings presented in this study need to be confirmed in an independent study with a larger study population that allows analysis based on ethnicity, socioeconomic, and educational backgrounds. Ultimately, the terminology used to express the diagnosis of nonepileptic events and the manner in which this is communicated will best be determined by the clinician based on his or her knowledge of the individual family.

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