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Original Article

A Novel Parent Questionnaire for the Detection of Seizures in Children

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ABSTRACT

BACKGROUND: We developed a seizure questionnaire that could be administered by a trained research assistant in a two-step process, approximating the clinical diagnostic process of a pediatric epileptologist. This questionnaire was designed to study seizure prevalence in a research population of 10-year-old children at risk for epilepsy. **METHODS:** English-speaking parents of children 6 months to 12 years old were recruited from the pediatric neurology clinics at Boston Medical Center and interviewed using a computerized questionnaire. An algorithm of parent responses rendered a 4-level ranking scale of seizure probability for events: (1) not likely, (2) indeterminate, (3) probable, (4) almost certain. Blinded to questionnaire results, pediatric neurologists served as the diagnostic gold standard, ranking each patient event using the same four-level scale based on clinical history and examination. **RESULTS:** The questionnaire was completed by 150 of 177 (84.7%) enrolled parents. Seizure prevalence among participants was 38.6%. The seizure questionnaire yielded a fitted receiver operating characteristic area of 0.93 (95% confidence interval [CI], 0.89-0.97). Based on optimal sensitivity and false-positive fraction, we dichotomized the questionnaire results as consistent with seizure (levels 3 and 4) or without seizure (levels 1 and 2). Overall, findings included a 91.4% sensitivity (95% CI, 84.2%-98.6%) and an 82.6% specificity (95% CI, 74.9%-90.4%). The positive predictive value was 76.8% (95% CI, 66.9%-86.8%) and the negative predictive value was 93.8% (95% CI, 88.6%-99.1%). **CONCLUSIONS:** This pediatric seizure questionnaire was both sensitive and specific for detecting clinically confirmed seizures. This tool may be useful to researchers and clinicians in screening large populations of children, decreasing the time and cost of added neurological assessments.

Keywords: seizure, epilepsy, survey, questionnaire, pediatric, parent, children, validation studies

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Introduction

Determining seizure prevalence in populations of children is time consuming and costly. Accurate estimates of seizure prevalence in the general population are important for epidemiologic studies, and researchers may require such estimates within specific populations at risk for epilepsy. However, the expert resources needed to diagnose a seizure clinically preclude accurate estimates in most circumstances. Moreover, pediatric neurologists are scarce in many geographic regions, and many children with spells concerning for seizures must wait months before a diagnosis can be made. At present, no instrument exists to screen children and determine the likelihood that a spell is a true seizure for research or clinical purposes.

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Defining and identifying seizures is not simple for the clinician, and is even less clear for the researcher. In 1993 the Commission on Epidemiology and Prognosis of the International League Against Epilepsy (ILAE) proposed a definition of an epileptic seizure (hereafter referred to simply as seizure) for the purpose of epidemiologic research. The excerpt pertinent to our study is as follows: "A clinical manifestation presumed to result from an abnormal…discharge of…neurons in the brain. The clinical manifestation consists of sudden and transitory abnormal phenomena which may include alterations of consciousness, motor, sensory, autonomic, or psychic events, perceived by the patient or an observer."[1] The current clinical definition of seizure remains essentially unchanged and still lacks an operationalized approach for making a seizure diagnosis in individuals or in populations for research purposes.[2]

In the absence of witnessed seizures or seizures captured on electroencephalography (EEG), the gold standard for making a clinical diagnosis of seizure is an evaluation by an experienced pediatric neurologist with specialized training in pediatric epilepsy, who attempts to identify the above-noted clinical features of events. In anticonvulsant clinical trials, researchers focus on subjects with treatment-resistant epilepsy and measure seizure burden with video EEG. In population-based epidemiologic research, however, seizure occurrence is less frequent, and case ascertainment of subjects with seizures often is less clear.

The 2011 ILAE report on the standards for epidemiologic studies and surveillance of epilepsy offered guidance for identifying seizures. They offered that the diagnosis should be made by a person with specialized training in epilepsy based on the presence of the clinical manifestations detailed above. Yet most epidemiologic research has relied on International Classification of Disease Codes, medical record reviews, and, less often, seizure screening instruments.[3-10]

To address the limitations in accurately identifying seizures, we developed a two-step questionnaire approximating the diagnostic algorithm of a trained expert clinician. Our aim in this report is to detail the development of a seizure diagnostic tool for children and to describe its reliability and validity.

Materials and Methods

Seizure survey development

We developed a seizure survey that incorporated the following considerations. First, in the absence of witnessed seizures or seizures captured on EEG, the gold standard for making a diagnosis of seizure is based on an experienced pediatric neurologist with specialized training in pediatric epilepsy. Second, the vast majority of children do not have seizures. As a result, our group of pediatric neurologists with specialized training in childhood seizures and epilepsy created a tool with two parts (online supplement). Because we planned to administer the completed instrument in a study of seizure prevalence in nearly a thousand 10-year-old children at risk for epilepsy, we structured the survey to be administered by a trained research assistant. Part 1, an 11-item questionnaire, was designed to be highly sensitive but less specific, allowing a subsequent, more in-depth evaluation to be limited to a smaller number of subjects. Screening questions included broad symptoms of seizure-like behavior, a prior history of seizures or epilepsy, or a history of having had an EEG in the past. Part 2, a 30-question panel, was designed as a comprehensive set of questions simulating the clinical interview and diagnostic schema used by an expert pediatric epileptologist to discern seizure from a nonepileptic event. Questions addressed possible ictal and postictal symptoms witnessed during the event. This 30-question panel was developed using a modified Delphi method, followed by alpha testing with caregivers to identify problems with intelligibility or question flow. The questionnaire was reviewed to ensure that the language was understandable at the eighth grade level (i.e., a Flesch–Kincaid grade level of 8.0). Questions were reviewed for content validity by one of the epileptologists not involved in the original development of the tool or in any other portion of its investigation. The logical process we applied was transformed into a computer-generated algorithm used to decide if an event was likely to be a seizure.

The intention of this questionnaire was to develop a research tool that could be used in populations in which detailed medical information, such as EEG results, may not be available. EEG, therefore, had a limited role in the seizure algorithm. If a child had an EEG in the past, this prompted the automatic administration of Part 2 of the seizure questionnaire. EEG results were not collected as part of the seizure questionnaire; however, because the treating neurologist rated the events, knowledge of previous EEG results may have contributed to their decision making.

In advance of testing the tool, we anticipated limitations in identifying some seizure types with this survey. Absence seizures may well escape detection as a seizure event on clinical grounds and requires confirmation with EEG. Focal seizures without loss of consciousness are often difficult to identify with certainty on clinical grounds; they can also be difficult to identify with EEG.[11] Our questionnaire centered on determining the presence or absence of seizure, but was not designed to determine specific epilepsy syndromes or seizure types.

In the planning of this study, we were concerned about possible recall bias. We attempted to minimize recall bias by designing similar questions that were repeated and rephrased, allowing parents more opportunities to reflect on past events. In addition, we recorded the age of the child at the time of the interview, the child's age at the onset of each event type, the frequency of each event type, and the time interval between the interview date and the last time each event type was observed.

Study subjects

Guardians of children ages 6 months to 12 years, followed in the Pediatric Neurology Clinic at Boston Medical Center for any neurological condition, were recruited by one of five treating pediatric neurologists, all with over 10 years of experience evaluating children for possible seizures. Consent was obtained from guardians by a research assistant. Following recruitment, patients were given a unique identification number to ensure confidentiality. The seizure questionnaire was designed to be completed by the care provider who spent the most time with the child and had the highest likelihood of witnessing an event. Although the study design allowed for the questionnaire to be administered to someone other than the legal guardian if identified as the primary caregiver, in each instance the guardian identified themselves or their coguardian. Only English-speaking guardians were recruited.

Interviewers

Interviewers had a college education and none were trained health professionals. To optimize administration reliability, interviewers were trained and then observed administering the survey to two or three subjects by a study coinvestigator. Caregivers had the choice to complete the questionnaire in person or at a more convenient time by telephone.

Conducting the seizure questionnaire

The two parts of the questionnaire were administered in person or by telephone interview. The interviewer logged into a computerized version of the questionnaire, which prompted the interviewer to the sequence of questions. The interview began with a standardized introductory script and was followed by Part 1 of the Questionnaire, a 2-3 minute screen.