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Development of the PedsQL™ Epilepsy Module: Focus group and cognitive interviews

Katherine W. Follansbee-Junger [a], Krista A. Mann [a], Shanna M. Guilfoyle [a], Diego A. Morita [b], James W. Varni [c], Avani C. Modi [a,*]

[a] Division of Behavioral Medicine and Clinical Psychology, Cincinnati Children's Hospital Medical Center, 3333 Burnet Ave., Cincinnati, OH 45229-3039, USA
[b] Division of Neurology, Cincinnati Children's Hospital Medical Center, 3333 Burnet Ave., Cincinnati, OH 45229-3039, USA
[c] College of Architecture, Texas A&M University, 3137 TAMU, College Station, TX 77843-3137, USA

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ABSTRACT

Youth with epilepsy have impaired health-related quality of life (HRQOL). Existing epilepsy-specific HRQOL measures are limited by not having parallel self- and parent-proxy versions, having a restricted age range, not being inclusive of children with developmental disabilities, or being too lengthy for use in a clinical setting. Generic HRQOL measures do not adequately capture the idiosyncrasies of epilepsy. The purpose of the present study was to develop items and content validity for the PedsQL™ Epilepsy Module.

Methods: An iterative qualitative process of conducting focus group interviews with families of children with epilepsy, obtaining expert input, and conducting cognitive interviews and debriefing was utilized to develop empirically derived content for the instrument. Eleven health providers with expertise in pediatric epilepsy from across the country provided feedback on the conceptual model and content, including epileptologists, nurse practitioners, social workers, and psychologists. Ten pediatric patients (age 4–16 years) with a diagnosis of epilepsy and 11 parents participated in focus groups. Thirteen pediatric patients (age 5–17 years) and 17 parents participated in cognitive interviews.

Results: Focus groups, expert input, and cognitive debriefing resulted in 6 final domains including restrictions, seizure management, cognitive/executive functioning, social, sleep/fatigue, and mood/behavior. Patient self-report versions ranged from 30 to 33 items and parent proxy-report versions ranged from 26 to 33 items, with the toddler and young child versions having fewer items.

Conclusions: Standardized qualitative methodology was employed to develop the items and content for the novel PedsQL™ Epilepsy Module. The PedsQL™ Epilepsy Module has the potential to enhance clinical decision-making in pediatric epilepsy by capturing and monitoring important patient-identified contributors to HRQOL.

1. Introduction

Youth with epilepsy have impaired health-related quality of life (HRQOL) compared with both healthy peers and those with other chronic medical conditions [1–3]. HRQOL is a multidimensional construct that measures patient perceived impacts of health across a range of dimensions, including physical, emotional, academic, and social domains [4]. In an era of increased attention to patient-reported outcomes [5], HRQOL is considered an important clinical measure in pediatric medical care [6]. Accordingly, being able to accurately capture this information in a developmentally-sensitive, valid, and expeditious way is essential for both clinical and research applications.

To date, the field of pediatric epilepsy has used generic HRQOL measures such as the PedsQL™ 4.0 Generic Core Scales [4], which are relatively brief (23 items) and beneficial when making comparisons across a wide variety of pediatric populations [7]. Conversely, generic measures may not fully capture the idiosyncratic impacts of epilepsy on functioning or be sensitive to changes in disease status [6]. This limits their utility as a disease-specific clinical outcome measure. Epilepsy-specific HRQOL measures, including the Quality of Life in Epilepsy-Adolescent-48 [8] and the Quality of Life in Childhood Epilepsy [9], capture a broader range of domains than generic measures. These measures focus on disease and treatment issues that are salient in epilepsy. However, several notable weaknesses compromise their usefulness. For example, the Quality of Life in Epilepsy-Adolescent-48 is a self-report measure for adolescents (ages 11–17) only, which is a significant limitation given that the median age of onset of childhood epilepsy is between

* Corresponding author at: Cincinnati Children's Hospital Medical Center, Division of Behavioral Medicine and Clinical Psychology, Center for Adherence and Self-Management, 3333 Burnet Ave., MLC 7039, Cincinnati, OH 45229-3039, USA. Tel.: +1 513 636 4864; fax: +1 513 803 0415.
E-mail address: avani.modi@cchmc.org (A.C. Modi).

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5 and 6 years [10]. In addition, no parent proxy-report version was co-developed with the Quality of Life in Epilepsy-Adolescent-48. Obtaining information from both parents and patients is optimal as they can offer unique and valuable perspectives about their subjective experiences [11]. The Quality of Life in Childhood Epilepsy (USA Version) instrument covers a wider developmental spectrum (parent report of 4–18 year olds) than the Quality of Life in Epilepsy-Adolescent-48; however, there is no companion self-report measure, and the measure is 79 items long. Lastly, both the Quality of Life in Epilepsy-Adolescent-48 and the Quality of Life in Childhood Epilepsy were developed without the inclusion of individuals with learning or intellectual disabilities. Considering that 1/3 of patients with childhood-onset epilepsy have intellectual or learning disorders [12] and recognizing its impact on HRQOL [13], eliciting input during the item development stage from these patients and their caregivers could potentially reveal additional areas for the development of a new epilepsy-specific HRQOL measure.

The primary aim of the current study was to create an epilepsy-specific module of the PedsQL™ that will build on existing measures in several important ways. First, our measure will assess a broad age spectrum of 2–18 years in a developmentally appropriate fashion. Second, self-report and parent proxy-report versions will be developed in tandem for children age 5–18. Third, the increased relevancy of the measure will lie in its ability to assess QOL in all youth with epilepsy and its increased sensitivity to factors impacting QOL in youth with cognitive and learning comorbidities.

The methodology to develop a PedsQL™ disease-specific module has previously been reported [14,15]. In line with these established procedures, development of the PedsQL™ Epilepsy Module has occurred in several phases: 1) content generation (Phase 1); 2) construction of the initial measure and item pool (Phase 2); 3) pretesting and instrument refinement (Phase 3), 4) national validation (Phase 4); and 5) dissemination and international translation (Phase 5). The purpose of the current paper is to describe the first three steps of the validation process, including how the items were generated, modified, and adapted based on a thorough literature review (Phase 1a), expert feedback (Phases 1a and 2b), focus groups (Phase 2a), and cognitive interviewing with children with epilepsy and their caregivers (Phase 3). Through this rigorous content validation process, a PedsQL™ Epilepsy Module will be created for national validation testing for children 2–18 years of age with epilepsy, with parallel pediatric patient self-report, and parent proxy-report versions.

2. Methods

2.1. Participants

Study participants were youth ages 5–18 years diagnosed with epilepsy and their primary caregiver, as well as primary caregivers of children age 2–4 years diagnosed with epilepsy. Families were recruited during routine medical visits through the Comprehensive Epilepsy Center at Cincinnati Children's Hospital Medical Center. Recruitment was targeted to ensure that our participants represented the spectrum of ages, developmental abilities, sex, and type of epilepsy (e.g., newly diagnosed, intractable, surgical patients, and well-controlled). Participants met the following inclusion/exclusion criteria: 1) child aged 2–18 years, 2) diagnosis of epilepsy, 3) ability to read and speak English due to the questionnaires only being validated in English, 4) no other major medical diagnoses, and 5) no autism spectrum disorders (ASDs). Caregivers of children with ASD were not included in this sample because we thought it would be difficult for caregivers to separate out cognitive and behavioral symptoms that were related to the ASD versus epilepsy and associated treatments. Notably, if children were reported to have developmental or language delays, parents could participate in the focus groups/cognitive interviews but the children did not participate.

2.2. Procedure

Potential participants and their caregivers meeting initial eligibility criteria were identified by a trained research assistant. A thorough overview of the study was provided, including study procedures, benefits, and risks. All questions were addressed, and informed consent/assent was obtained. All study procedures were approved by the hospital's Institutional Review Board.

2.2.1. Construction of the initial measure and item pool

2.2.1.1. Content generation, construction of the initial measure and item pool (phases 1 and 2). PubMed and Google Scholar were used to conduct a comprehensive literature review to generate content and develop the conceptual framework for the focus groups. Items for the measure were developed through an iterative process including literature review, expert input, focus groups, and cognitive interviewing. Cognitive interviewing is a commonly used procedure in measurement development designed to identify problems with item comprehension, recall, and other cognitive processes that can be modified by rewording or reordering items on a measure. Using a think aloud procedure, caregivers and patients are asked to verbalize their thoughts and responses on the measure with prompting by the interviewer [16,17]. An item bank was developed from existing measures focused on generic HRQOL, epilepsy-specific HRQOL, and the PROMIS database. The items included in the PedsQL™ Epilepsy Module were developed to capture the impact of epilepsy symptoms and treatments on patient and family functioning based on the perspectives of parents, youth with epilepsy, and experts involved in their care. Each parent and child version of the PedsQL™ Epilepsy Module was consistent with the instructions, time frame, format, and answer choices of the generic PedsQL™.

2.2.1.2. Focus groups. Focus groups were conducted by licensed psychologists and a graduate student. Semi-structured, open ended questions were asked of the participants to identify and develop content items. Participants were asked to discuss how epilepsy and/or its treatments affect functioning in physical activities, social interactions, mood, behavior, learning and academics, and family life. Parents and children age 8–18 years participated in separate simultaneous focus groups. Children age 5–7 years participated in short interviews with their parents present, and then participated in recreational activities while parents completed their portion. Parents of children age 2–4 years participated without their child. Six focus group sessions were transcribed based on audio and video-recordings. These six transcripts (3 child and 3 parent focus group) were coded by two independent reviewers (SG & JV) for thematic content. Parent and participant responses were separated and then grouped according to age and subject. Three researchers (KJ, KM, & AM) examined the thematic content, and final decisions were made by consensus. Thematic saturation was achieved, that is families were not identifying new content by the end of all focus groups.

Initial child self-report and parent proxy-report versions were developed to encompass the themes elicited during the literature review, expert input, and focus group interviews. The draft versions were sent out to epilepsy experts across the country for additional feedback.

2.2.2. Pretesting and instrument refinement (phase 3)

2.2.2.1. Cognitive interviewing. In the cognitive interviewing phase, the updated draft of PedsQL™ Epilepsy Module was reviewed by a unique cohort of children with epilepsy and parents of children with epilepsy who were not participants in the previous focus interviews. Both cohort groups were divided among the patient age ranges of 2–4, 5–7, 8–12, and 13–18 years of age, consistent with previous PedsQL™ age groupings. Participants completed the PedsQL™ Epilepsy Module and then provided feedback employing the previously described respondent debriefing methodology [16,17]. The goal of the cognitive interviews