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Response shift in parents' assessment of health-related quality of life of children with new-onset epilepsy

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ABSTRACT

Objective: Diagnosis of epilepsy is known to impact health-related quality of life (HRQOL) of children with new-onset epilepsy and can also influence their conceptualization and valuation of HRQOL construct, also known as response shift. This study investigates the presence of response shift in a cohort of children with new-onset epilepsy.
Methods: Data are from the HEalth-Related QUality of Life in children with Epilepsy Study, a prospective cohort study of 373 children with new-onset epilepsy. Hypotheses about the presence of reconceptualization, reprioritization, and recalibration response shift were tested in the Quality of Life in Childhood Epilepsy (QOLCE-55) Questionnaire, a parent-reported, disease-specific HRQOL measure, using Oort's structural equation model between baseline and 1-year follow-up. Model fit was assessed using log-likelihood ratio test, root mean square error of approximation, and comparative fit index.
Results: Small positive uniform recalibration response shift effects were observed on physical, emotional, and social functioning domains of the QOLCE-55, but negligibly small negative nonuniform recalibration response shift effect was observed on social functioning domain. There was no significant change in overall QOLCE-55 scores over time after adjusting for response shift effects.
Significance: Parents of children with new-onset epilepsy are likely to positively recalibrate (upward bias) their assessments of their children's HRQOL over a 1-year period after diagnosis. This study highlights the potential benefits of response shift as a desired consequence in parents' perception of changes in HRQOL of children with new-onset epilepsy.

1. Introduction

The diagnosis of epilepsy has a significant impact on patients' clinical, psychosocial, and health-related quality of life. The diagnosis of epilepsy during childhood (the most common period of disease onset) also impacts the lives of the children's families in many ways, including the burden of clinical care, limited social interactions due to unpredictability of epilepsy seizures [1], and family finances as parents may spend less time or no time in paid employment to care for their child's epilepsy [2,3]. The diagnosis of epilepsy may also result in psychiatric, behavioral, and cognitive comorbidities, with consequent long-term negative effects [4,5]. The goal of epilepsy management in these children is sustained seizure control and consequently decreased disease burden and improvement in patients' health-related quality of life (HRQOL). Since sustained seizure control does not consistently improve HRQOL, the accurate measurement and interpretation of changes in HRQOL are important in children with new-onset epilepsy [6].

However, there is increasing awareness that the assessment and interpretation of longitudinal change in HRQOL may be confounded by response shift, a change in patients' perception of their health and well-being [7,8]. Response shift occurs in patient- or proxy-reported outcomes when a catalyst (e.g., treatment intervention, disease diagnosis) triggers a shift in an individual's perception of his/her health status and well-being [7]. According to Sprangers and Schwartz [8], response shift is defined as a change in an individual's internal standards (recalibration), values (reprioritization), or conceptualizations (reconceptualization) of the target

Abbreviations: CFI, comparative fit index; ES, effect size; HRQOL, health-related quality of life; HERQULES, Health-Related Quality of Life in Children with Epilepsy Study; QOLCE, Quality of Life in Childhood Epilepsy; QOLCE-55, 55-item Quality of Life in Childhood Epilepsy; RMSEA, root mean square error of approximation.

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construct (e.g., health status or quality of life). Recalibration response shift occurs when an individual's interpretation of the measurement scale changes over time. For example, patients with new-onset epilepsy may initially rate their illness as very severe; however, over time, they may develop strong coping mechanisms and adapt to their disease in such a way that they report better HRQOL, even though their clinical symptoms have not necessarily improved. Reprioritization response shift arises when there is a change in an individual's values toward the health construct; it manifests through overall changes in the importance an individual attributes to different health-related quality of life domains (e.g., physical, mental and social health domains) that are relevant to the overarching target construct. Reconceptualization response shift occurs when individuals tend to redefine the target construct the construct itself over time (e.g., new domains may emerge). A new set of measurement indicators for the construct is needed to ensure that the full scope of the intended target construct is adequately represented. While much has been written about the presence and detection of response shift effects in longitudinal studies of patient-reported outcomes in general, there is limited investigation of response shift effects in epilepsy, and it is scarce in pediatric chronic diseases like epilepsy. This may be partly attributed to the reliance on proxy reports (e.g., caregivers, parents, and clinicians), which are extensively used to assess HRQOL in children with epilepsy [9]. Importantly, proxy reports are generally considered to be less sensitive to detect response shift effects than patients' direct reports, mostly because proxy raters do not experience the diagnosis themselves [10,11]. Hence, moderate to smaller response shift effects are usually reported in proxy reports.

In this study, we investigate the presence of response shift in parent-reported children's HRQOL, as measured by the Quality of Life in Children with Epilepsy (QOLCE-55) Questionnaire over a 1-year period. We postulate that the diagnosis of epilepsy is a catalyst that triggers response shift in these patients and that small to moderate effect sizes will be observed in parent-reported children's HRQOL in this cohort.

2. Methods

2.1. Data source and study sample

Data for this study are from The Health-Related Quality of Life in Children with Epilepsy Study (HERQULES), a 2-year prospective cohort study assessing the course and determinants of HRQOL in children with new-onset epilepsy across Canada [12]. Using a two-stage clustered sampling strategy between April 2004 and April 2007, 53 (74%) of practicing pediatric neurologists in Canada recruited parents of children with epilepsy (median: 9 families per neurologist). The sample included children ages 4 to 12 years with ≥2 unprovoked new-onset seizures. The children were seeing a pediatric neurologist for the first time and had not received confirmation of the diagnosis of epilepsy previously. Details of HERQULES have been described elsewhere [13]. Data were collected at baseline (as close to the time of diagnosis of epilepsy) and at approximately 6 months (time 2), 12 months (time 3), and 24 months (time 4) later. Standardized questionnaires were used to collect parent report of their children's HRQOL and a series of child and family characteristics, while a neurologist-report form collected information on clinical characteristics of the child's epilepsy. Ethical approval for HERQULES was obtained from all relevant research ethics boards across the country.

2.2. Quality of Life in Childhood Epilepsy (QOLCE-55)

The QOLCE is a parent-reported epilepsy-specific scale for evaluating the HRQOL of children aged 4–18 years. The original version of the questionnaire consists of 76 items with 16 subscales covering 7 domains of life function [14]. The QOLCE-55 is a recently modified version of the original 76-item QOLCE15 [15], and it measures 4 HRQOL domains: cognitive functioning (22 items), emotional functioning (17 items), social functioning (7 items), and physical functioning (9 items). Each item uses a 5-point Likert scale as follows: 0 = very often; 1 = fairly often; 2 = sometimes; 3 = almost never; and 4 = never. Ratings undergo linear transformation such that domain scores can take values from 0 (low HRQOL) to 100 (high HRQOL). The QOLCE-55 has high internal consistency ($\alpha = 0.96$), reliability, and measurement equivalence [15,16]. The QOLCE-55 was administered at each occasion.

2.3. Statistical analysis

Descriptive analysis of means, standard deviation, and frequency distributions were used to summarize patients' characteristics at baseline. Repeated-measures multivariate analysis of variance was used to assess longitudinal changes in each of the QOLCE-55 domains between baseline and 1-year follow-up for children with epilepsy.

We tested for the presence of response shift among the domains of QOLCE-55 using Oort's proposed four-step procedure based on structural equation models [17,18]. Specifically, changes in intercepts indicate uniform recalibration, changes in residual variances indicate nonuniform recalibration, changes in the patterns of common factor loadings indicate reconceptualization (i.e., a factor loading goes from zero to nonzero), and changes in common factor loadings indicate reprioritization. In Step 1, a measurement model is fit to the data without any across-occasion constraints, with all the model parameters allowed to vary over time. In Step 2, a model that assumes equivalence of the latent constructs over the 1-year period (i.e., no response shift model) is fit to the data by imposing equivalence constraints on intercepts, factor loadings, and residual variances over time.

The adequacy of the models in each step was assessed using three fit indices, namely, χ^2 goodness-of-fit test, root mean square error of approximation, and the comparative fit index (CFI). For RMSEA, a value of 0.05 or less is indicative of close model fit, a value between 0.05 and 0.08 is indicative of reasonable fit, and a value of 0.10 or greater is indicative of poor fit [19–21]. For CFI, a value greater than 0.90 suggests reasonable fit. Moreover, changes in fit indices across the steps will be used to establish the presence of response shift. Statistical significance based on changes in χ^2 goodness-of-fit test and practical significance based on changes in CFI and RMSEA will be used for model comparisons. Given that the χ^2 goodness-of-fit test is known to be sensitive to sample size [18,21], evidence about statistical or practical significance from at least two of the three fit indices will be used to establish evidence against the null hypothesis that there is no response shift. Consistent with previous reports in the literature, changes in RMSEA and CFI greater than 0.015 between measurement models are considered significant.

Fit indices are used to compare the models in Step 1 and 2. Response shift is considered not present if the change in at least two of the three fit indices is not significant, and no further analyses will be conducted. Otherwise, we proceed to Step 3, in which the constraints in Model 2 (fully constrained models) are sequentially removed starting with the largest modification index to improve the fit of the model from Step 2. Each modification is tested using a likelihood ratio statistic. The modification steps are mainly guided by Lagrangian multiplier tests (i.e., modification indices), expected parameters of change, Wald tests, and the inspection of standardized residuals [17]. In Step 4, the final step, tests of specific across-occasion invariance constraints in common factor means, factor variances, and factor correlations are applied to the data [18]. Differences in common factor means are indicative of true change in the means, while differences for common factor variances are indicative of true change in the variances. Differences for common factor residuals are indicative of both reconceptualization and reprioritization. The estimated, standardized true changes are then interpreted according to Cohen's guidelines for measures of effect sizes [22].

Additional analyses were conducted to examine the influence of covariates such as number of antiepileptic drugs, cognitive problems,