Preliminary Reliability & Validity of the PedsQL Family Impact Module in Families of Children With Barth Syndrome: Using Propensity Score Matching

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PURPOSE: Barth syndrome (BTHS) is a rare, X-linked disorder found primarily in boys. This study examined the preliminary reliability and validity of the PedsQL Family Impact Module (PedsQL FIM) in families of children with BTHS.

DESIGN: In this cross-sectional study, 33 parents of children with BTHS (BTHS group) and 39 parents of typically developing children (control group) participated.

METHOD: Both groups of parents completed the PedsQL FIM, which assesses parental health-related quality of life (HRQoL) and family functioning, and a demographic information form. Internal consistency reliability and item-total correlations were calculated to test the reliability of the PedsQL FIM. Construct validity was examined using the known-groups method. We estimated the mean score differences of the PedsQL FIM between the two groups using three different models, including unadjusted, multivariate regression, and propensity score matching with inverse probability of treatment weighting (PS-IPTW) models.

RESULTS: The Cronbach's alpha coefficients were greater than 0.70 for all scales of the PedsQL FIM, except for the Communication scale in the BTHS group. The item-total correlations were significant for all scales with moderate to high correlations ($p < .05$). In construct validity, the mean scores of the PedsQL FIM between the two groups were significantly different ($p < .05$) for all scales and total score in the unadjusted and PS-IPTW models.

CONCLUSION: The PedsQL FIM demonstrated adequate measurement properties of preliminary reliability and validity in assessing the impact of children with BTHS on parental HRQoL and family functioning.

IMPACT STATEMENT: Measuring family impact using a reliable and valid instrument, the PedsQL FIM, will help better understand the impact of raising children with BTHS on families. It will also provide crucial information for occupational practitioners to implement holistic and comprehensive healthcare services for families.

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