Abstract

Introduction. The objective of this study was twofold: first, to conduct a confirmatory factor analysis (CFA) of the Portuguese versions of Disabkids-10; and second, to examine potential differences in factor structures between age groups, genders and informants.

Method. The sample included 293 school-aged children and adolescents with chronic health conditions and 197 parents. Both family members (whenever possible) completed the self- and proxy-report versions of Disabkids-10.

Results. The factorial model of Disabkids-10 had good fit for self-reported data and minimally acceptable fit for proxy-reported data. The multi-group analyses confirmed the model invariance across age groups (children vs. adolescents), genders (boys vs. girls) and informants (children vs. parents).

Discussion. The generic developmental applicability of these questionnaires makes them recommended for healthcare routine assessments on pediatric intervention needs and outcomes.
Introduction

During the last couple of decades, in a context of elevated prevalences and improved survival rates for a number of chronic health conditions, health-related quality of life (HRQL) assessment has become increasingly important in pediatric health care research and practice (Clarke and Eiser, 2004). This redefinition of health outcomes was motivated by a need of complementing simple biological or physiological endpoints (e.g., growth, mortality), with more meaningful outcomes of child and adolescent care and development (Christakis et al., 2001). Alongside the fact that many clinicians tend to regard pediatric HRQL measurement as too costly or impractical (Varni et al., 2005), there has been a growing demand for assessing HRQL more economically (Muehlan, 2010). On the other hand, despite the acknowledgement of differences between age groups, genders and informants in HRQL assessments (Bullinger et al., 2006; Wallander et al., 2001), measures focusing on common HRQL markers and allowing completion by both patients and proxies (usually parents) are certainly valuable in facilitating data collection and analysis. Therefore, a crucial research question that needs to be addressed is whether brief generic instruments are cost-effective methodologies in providing reliable HRQL measurements for pediatric patients of both genders and different age groups, as well as across informants.

HRQL is “a multidimensional concept that includes the broad areas of functional status, psychological and social well-being, health perceptions, and disease- and treatment-related symptoms” (Aaronson et al., 1991: 840). The concept of HRQL incorporates a medical and healthcare perspective and is thus a component of the more general notion of quality of life, which integrates a wider range of aspects such as political freedom and financial issues (Koot, 2001; The European Disabkids Group, 2006). Clinical rationales for assessing HRQL include the identification of dysfunction secondary to illness or treatment, the delineation of patient subpopulations at risk for poor psychosocial outcomes, the
evaluation of the quality of medical care, the improvement of clinical decision making, and the estimation of healthcare needs of a given population (Spieth and Harris, 1996). In fact, HRQL assessments can predict costs of care for pediatric populations and assist the management of care by enhancing clinical evaluations and identifying suitable candidates for case management (Seid et al., 2005). In this context of routine monitoring and screening, brief measures that summarize scores into a single value (or index) may be especially useful for examining global changes in HRQL, while reducing response burden and saving administration costs (Ravens-Sieberer et al., 2010).

In pediatric healthcare settings, developmental issues are pervasive challenges for HRQL assessment: not only there are reciprocal effects of chronic illness/disability and child/adolescent development (Suris et al., 2004), but also development affects the selection of targeted dimensions, item content, instrument format and the use of proxy information (Koot and Wallander, 2001). Therefore, a developmental approach to HRQL assessment calls for the integration of age-related specificities and the consideration of parents’ and their children’s reports as complementary to each other (Bruil and Detmar, 2005; Carona et al., 2012). Despite the overall applicability of three broad domains of HRQL (i.e., physical, psychological and social) to children and adolescents, there may be substantial variation in their content between age groups and genders (Bullinger et al., 2006; Rajmil et al., 2004). Compared to children, adolescents seem to report fewer positive emotions, be more mindful of others’ opinions, have more varied social activities and consider more the distant than the immediate future. Gender differences in HRQL meaning, in contrast, tend to be fewer and to mostly occur at the item level of specific activities (e.g., boys are more likely to describe sports and computers activities, and girls other activities such as dancing, shopping and chatting on the phone) (Wee et al., 2006). Nevertheless, many HRQL studies cover a wide age range with no stratification of results (Gerharz et al., 2003), even if such aggregation across age groups and genders may obscure important differences. On the topic of multiple informants in pediatric HRQL assessment, it has been commented that both parents’ and their children’s HRQL reports
are valid, although they cannot effectively supplant each other (Theunissen et al., 1998). If on the one hand, proxy reports that are usually provided by parents, may be the only way to evaluate HRQL in some patients, due to their young age, severity of illness/disability or cognitive impairment (White-Koning et al., 2007), on the other hand, children and adolescents’ self-reports are essential to incorporate a child’s perspective within healthcare care settings and thus promote a truly child-centred care (Söderbäck et al., 2011; Varni et al., 2005). For these reasons, it is necessary to ensure that self and proxy-report versions of a given HRQL instrument accurately assess the same construct across different informants.

The Disabkids questionnaires represent a sound methodology to assess pediatric HRQL in a developmentally appropriate way and include self and proxy-report forms for a chronic generic module (long and short versions), seven condition-specific modules (e.g., diabetes, epilepsy, dermatitis), and a measure of Smiley for younger children. The short version of the chronic generic module, known as Disabkids-12 or Disabkids-10+2, was developed to assess pediatric HRQL in a more economic way and it was based on the conceptual model of three higher-order domains (Mental, Social, Physical) and six facets (Independence, Emotion, Inclusion, Exclusion, Limitation, Treatment) that underlay the longer version of the generic module, known as Disabkids-37 (Muehlan, 2010; The European Disabkids Group, 2006). Although the short version of the instrument initially included two items per each one of the aforementioned facets, later studies indicated a preferred use of an index score based on 10 items, thus excluding the “applicable” items derived from the “Treatment” facet, and allowing the completion of questionnaires by the generality of pediatrics patients, whether they were undergoing medical treatment or not. The measurement model was then examined for this one-factor solution via confirmatory factor analysis (CFA), and results indicated good and acceptable model fits for the self and proxy versions of Disabkids-10, respectively (Muehlan, 2010). Nevertheless, the invariance of such one-dimensional measurement model across age groups, genders and informants remained to be ascertained.
The question of whether a pediatric HRQL measure indeed measures that target construct in a given population is intrinsically related to the notion of construct validity. Construct validity is at the heart of psychometric assessment and may be defined as “the extent to which an instrument can provide a good representation of a construct” (Wallander, 2001: 25). A widely used method to investigate construct validity is CFA, in which, contrary to exploratory factor analysis, factor structures are hypothesized a priori and then empirically verified rather than extracted from the data (Lei and Wu, 2007). CFA increases the statistical precision on the investigation of construct validity by reducing measurement error and assisting the comparison of factors structures between two or more groups (Atkinson et al., 2011).

Using this statistical procedure, the aim of the present study was twofold: first, to conduct a CFA of the Portuguese versions of Disabkids-10 in a mixed pediatric sample; and second, to ascertain the invariance of Disabkids-10 measurement model across age groups (children vs. adolescents), genders (boys vs. girls), and informants (parents vs. children).

Method

Participants and Procedures

The sample was composed of 293 pediatric patients, between 8 and 18 years-old, with three different clinical diagnoses, and 197 parents/family caregivers. The sample comprised cases of diabetes, epilepsy and cerebral palsy, because these three chronic health conditions were already included in the original studies of Disabkids questionnaires (Muehlau, 2010; The European Disabkids Group, 2006). The selection of these conditions was also thought to provide an interesting and varied sample of pediatric patients, as suggested by the following evidence: firstly, type 1 diabetes is one of the most common chronic diseases in children, for which an increasing prevalence has been reported (Passa, 2002);
secondly, epilepsy is among the most prevalent neurological conditions in the developing years (Ronen et al., 2003); and lastly, cerebral palsy is the most common physical disability in childhood (Stanley et al., 2000) and has been described as an interesting prototype of childhood disability (Raina et al., 2004). The participants’ socio-demographic and clinical characteristics are presented in Table 1.

Using the non-probabilistic convenience sampling method, participants were selected between March 2009 and September 2012 in the outpatient services of four Portuguese public hospitals and 10 Portuguese Cerebral Palsy Associations (tertiary healthcare institutions), after the study had been approved by the institutions’ Ethic Committees and Direction Boards. For inclusion in the sample, pediatric patients had to meet the following inclusion criteria: (1) age between 8 and 18 years-old; (2) diagnosis of type 1 diabetes, epilepsy or cerebral palsy, established by a physician according to the International Classification of Diseases (ICD-10); (3) ability to understand and answer the questionnaires (for pediatric patients with cerebral palsy, data from previous formal assessment of their intelligence quotient [IQ] was collected and a value of 70 was set as the threshold); and (4) be accompanied by one of the parents or other family caregiver.

Informed consents were obtained from all parents and adolescents older than 13, and informal assents were obtained from children. The questionnaires were completed by pediatric patients and parents/family caregivers in a room provided for research purposes in the institution they attended, under the supervision of a trained research assistant. Since patients with diabetes were participating in a concurrent research project that did not include parents’ reports on their children’s HRQL, proxy-reports were then not obtained for this clinical group.

Measures
The DISABKIDS Chronic Generic Measure – Short Version assesses the HRQL of pediatric patients with chronic health conditions aged between 8 and 18 years-old (The DISABKIDS Group Europe, 2006). This short version comprises 10 items measuring the mental, social and physical impact of the health condition (e.g., “Does your condition get you down?”/ “Does your child’s condition get him/her down?”), and two items addressing the impact of treatment (e.g., “Does taking medication bother you?”/ “Does taking medication bothers your child?”). Following previous recommendations on the factorial examination of the measure (Muehlhan, 2010), and since the present study was aimed at testing an instrument that would apply to all pediatric patients, whether they were medicated or not, the two items assessing the impact of treatments were excluded from this study. The questionnaire reports to the “past four weeks” and is to be answered within a 5-point Likert scale ranging from 1 (never) to 5 (always), with higher scores indicating better HRQL.

Socio-demographic and clinical data were obtained from parents’ questionnaires.

**Data analysis**

Statistical analyses were performed with the Statistical Package for the Social Sciences (SPSS, v. 20; Chicago, IL, USA) and with the Analysis of Moments Structures (AMOS, v. 20). Missing data, that were random and lower than 5%, were handled by individual mean score substitution, except for socio-demographic and clinical variables.

To examine the factorial structure of Disabkids-10, first-order confirmatory factor analyses (CFA) were performed for both patient- and proxy-reported versions. The assessment of model’s fit was then based on (a) the significance and strength of factors loadings, and (b) the degree to which the overall model fitted the observed data, as suggested by a variety of indices (Weston and Gore, 2006).

Specifically, the overall models’ fit was assessed by examining the maximum-likelihood \( \chi^2 \) and the main approximate goodness-of-fit indexes, namely the comparative fit index (CFI) and the root mean square
error of approximation (RMSEA). A model was considered to have a good fit when $\text{CFI} \geq .95$ and $\text{RMSEA} \leq .06$; and was considered to have an acceptable fit when $\text{CFI} \geq .90$ and $\text{RMSEA} \leq .10$ (Browne and Cudeck, 1993; Hu and Bentler, 1999). Complementarily, a $\chi^2/\text{df}$ ratio of 5 or less was assumed as indicative of acceptable model fit (Ullman, 1996). In addition, construct reliability was assessed by using the Cronbach’s alpha value and the composite reliability (CR) value, calculated from the squared sum of standardized factor loading divided by the squared sum of standardized factor loading plus the sum of the error variance terms. Good construct reliability was established if CR was higher than .70 (Hair et al., 2010).

The factorial invariance of the models across age groups, genders and informants was tested in two steps, starting with the examination of the baseline model for each group separately, followed by multi-group analyses comparing the unconstrained model with a model in which factor loadings were fixed equal across groups (Byrne, 2010). The model invariance was established when the chi-square difference ($\Delta \chi^2$) was non-significant, and the difference in CFI values ($\Delta \text{CFI}$) was lower than .01 (Cheung and Rensvold, 2002).

**Results**

**CFA for the Self-report Version of Disabkids-10**

In order to examine the one-dimensional structure of the Disabkids-10, an initial model was tested, in which the 10 items were hypothesized to load on a single factor representing HRQL. According to the reference values for the main fit indexes, this model presented a marginally acceptable fit to the patient-reported data, with $\chi^2(35) = 139.50$, $p < .01$; $\chi^2/\text{df} = 3.99$; CFI = .86 and RMSEA = .10. Based on modification indices and on item content, the initial model was modified, by allowing the measurement
error terms for the items that belonged to the same facet in the long version of the questionnaire to correlate. This modified model, which is depicted in Figure 1, had a significantly better fit than the initial model, $\Delta \chi^2(5) = 76.00, p < .01$, improving the model fit indexes, $\chi^2(30) = 63.50, p < .01; \chi^2/df = 2.12; CFI = .96$ and RMSEA = .06. All of the items showed factorial validity, with statistically significant factor loadings ($p < .001$) and, except for items 3 and 10, standardized regression weights above the threshold of .50. The examination of Cronbach’s alpha and composite reliability values ($\alpha = .83; CR = .82$) confirmed that the 10 patient-reported items consistently represent the latent construct of HRQL.

CFA for the Proxy-report Version of Disabkids-10

Because we aimed to test a strictly equivalent model for both versions of the instrument, the factorial model that was modified to better fit the patient-reported data was also tested for the proxy-reported version. The modified model (Figure 2) had a minimally acceptable fit to the proxy-reported data, with $\chi^2(35) = 101.60, p < .01; \chi^2/df = 3.39; CFI = .93$ and RMSEA = .11, even though all of the items showed factorial validity with factor loadings above .50 and statistically significant. The proxy-reported questionnaire also presented good construct reliability, with Cronbach’s alpha = .90 and CR = .90.

Multi-group Analyses for Age Groups, Genders and Informants

To ascertain whether the Disabkids-10 Index was a valid measure to assess HRQL across different groups, the final factorial model was comparatively tested across age groups and genders. For the patient-reported data (Table 2), results showed that the model had acceptable fit for both age groups and genders, and multi-group analyses confirmed the invariance of factor loadings across groups.
For the proxy-report version (Table 3), the model presented poor fit for both gender groups and for children and acceptable fit for adolescents; the multi-group analyses showed no significant differences on factor loading between children and adolescents and between boys and girls, thus confirming the factorial validity across age groups and genders.

Finally, we tested for model invariance across informants (parents vs. children). The multi-group analyses confirmed the measurement invariance, $\Delta \chi^2(9) = 13.94, p = .12; \Delta CFI = .004$, i.e., that the factor loadings did not differ between patient- and proxy-reported data.

**Discussion**

This was the first study to investigate the invariance of a one-dimensional factor structure for Disabkids-10 questionnaires across age groups, genders and informants. The observed results add critical evidence for the overall applicability of Disabkids brief generic module to assess HRQL in pediatric patients with chronic health conditions, regardless of their age group and gender, or even of the selected informant for report.

In our first cluster of results, the proposed factorial model of Disabkids-10 displayed good and minimally acceptable fits for self and proxy-reported data, respectively. These results were remarkably similar with those originally reported in the European study of the instrument (Muehlan, 2010). A possible explanation for that discrepancy in model fits relies on the fact that Disabkids items were
formulated as a result of focus groups that primarily attended to children and adolescents’ perceptions (The European Disabkids Group, 2006). Consequently, the poorer goodness-of-fit indices observed for the proxy-report version do not discard its applicability in pediatric assessments, but instead imply the recommendations that children’s subjective self-reports should be considered whenever possible and parents/caregivers should be selectively used as proxies (White-Koning et al., 2007; World Health Organization, 1993).

In a second moment, our results further demonstrated the invariance of the one-dimensional measurement model for school-aged children and adolescents of both genders. In terms of construct validity assessment, this is to say that the items of Disabkids-10 reliably depict a general HRQL factor for pediatric patients of a wide age range, who are expectably facing different developmental contexts and dealing with distinct age-related tasks and challenges. Actually, even if major HRQL domains are developmentally universal, substantial variation may exist between age groups and genders at the level of instrument items (Rajmil et al., 2004; Wee et al., 2006). For this reason, despite the fact that Disabkids-10 items were extracted from the longer version of the instrument, for which semantic validation had previously established the developmental adequacy (Carona et al., 2011; The European Disabkids Group, 2006), factor analyses at the item level were crucial for determining the instrument’s construct validity in a developmentally appropriate way. Additionally, the invariance of Disabkids-10 measurement model was also ascertained across self and proxy-reported data, thus highlighting the instrument’s construct validity across informants. This result is particularly important because there are certain pediatric patients (e.g., patients severely injured and/or undergoing complex treatments, younger children, patients with cognitive impairment), whose HRQL assessments will mostly rely on their parents’ (or other proxies) reports.

Taken altogether, these results represent a vital contribution for the refinement of a developmental approach to HRQL assessment, which acknowledges age and gender specificities and
Therefore, distinctive strengths of the present study include: the utilization of CFA as a sophisticated statistical procedure to examine construct validity; the consideration of children and adolescents as independent age groups; and the simultaneous examination of self and proxy-report forms of a single pediatric HRQL measure. Nevertheless, three important limitations should be also acknowledged: first, although standing as a common psychometric procedure (cf. Muehlan, 2010), this validation study was based on data from the longer version of the instrument, thus requiring its future replication in an independent sample; second, proxy-reports were not obtained for all the clinical groups included in the study; third, the psychometric property of responsiveness to treatment over time, which is of paramount importance for pediatric settings (Varni et al., 2005), was not investigated; and last, although adequate for the intended purpose, this study’s sample comprised three distinct conditions, which even so did not comprehensively depict the most common pediatric populations. For this reason, future psychometric analyses of Disabkids-10 should be performed with data obtained from larger clinical samples, with a wider variety of chronic health conditions, in order to reliably assess the instrument’s applicability to different clinical diagnoses.

This study substantiated the utility of Disabkids-10 questionnaires to assess pediatric HRQL in a more economic way. In fact, these instruments comply with all the guidelines suggested by Clarke and Eiser (2004) to evaluate the quality of HRQL measures for pediatric practice or clinical trials, namely: the instrument’s reliability, validity and developmental appropriateness for the target group; the instrument’s briefness (i.e. containing less than 30 items); and the inclusion of self and proxy-report forms. The utilization of Disabkids-10 for routine assessment may be a practical way of incorporating HRQL measurement in clinical practice, which may ultimately assist the identification of hidden morbidities, facilitate patient-clinicians communication and clinical decision-making, and improve patient’s outcomes and satisfaction (Varni et al., 2005). Nevertheless, an index measure such as Disabkids-10 is inadequate
to obtain more detailed profile assessments, and its use should be complemented whenever possible with supplemental assessments that account for the most age-specific aspects (e.g., adolescent dating). Finally, future prospects for research on Disabkids-10 include the examination of the instrument’s responsiveness to medical or psychosocial treatments, as well as its performance as a screener in pediatric healthcare practice.
References


Table 1. Socio-demographic and clinical characteristics of the sample.

<table>
<thead>
<tr>
<th></th>
<th>Pediatric patients (n = 293)</th>
<th>Parents/caregivers (n = 197)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Socio-demographic characteristics</strong></td>
<td></td>
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<tr>
<td>Age, $M (SD)$</td>
<td>12.46 (2.96)</td>
<td>41.39 (6.56)</td>
</tr>
<tr>
<td>Age group, n (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Children 8-12</td>
<td>144 (49.1%)</td>
<td></td>
</tr>
<tr>
<td>Adolescents 13-18</td>
<td>148 (50.5%)</td>
<td></td>
</tr>
<tr>
<td>Missing</td>
<td>1 (0.3%)</td>
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<tr>
<td>Gender, n (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>146 (49.8%)</td>
<td>22 (11.2%)</td>
</tr>
<tr>
<td>Female</td>
<td>145 (49.5%)</td>
<td>174 (88.3%)</td>
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<tr>
<td>Missing</td>
<td>2 (0.7%)</td>
<td>1 (0.5%)</td>
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<tr>
<td>SES $^a$, n (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>113 (57.4%)</td>
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</tr>
<tr>
<td>Medium</td>
<td>52 (26.4%)</td>
<td></td>
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<tr>
<td>High</td>
<td>16 (8.1%)</td>
<td></td>
</tr>
<tr>
<td>Missing</td>
<td>16 (8.1%)</td>
<td></td>
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<tr>
<td><strong>Clinical characteristics</strong></td>
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</tr>
<tr>
<td>Diagnosis, n (%)</td>
<td></td>
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</tr>
<tr>
<td>Diabetes</td>
<td>96 (32.8%)</td>
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<tr>
<td>Epilepsy</td>
<td>104 (35.5%)</td>
<td></td>
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<tr>
<td>Cerebral palsy</td>
<td>93 (31.7%)</td>
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</tr>
<tr>
<td>Using medication, n (%)</td>
<td>211 (72%)</td>
<td></td>
</tr>
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</table>

$^a$ Socioeconomic status was determined using a classification system for the Portuguese context based on the parents’ jobs and educational levels (Simões, 1994).
Table 2. Factorial model comparison across age groups and genders for the self-report version of DISABKIDS-10.

<table>
<thead>
<tr>
<th></th>
<th>Model’s goodness of fit</th>
<th>Multi-group analyses</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$\chi^2$ (30)</td>
<td>$p$</td>
</tr>
<tr>
<td><strong>Age groups</strong></td>
<td></td>
<td></td>
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<tr>
<td>Children 8-12</td>
<td>50.31</td>
<td>.012</td>
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<tr>
<td>Adolescents 13-18</td>
<td>59.11</td>
<td>.001</td>
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<tr>
<td><strong>Gender</strong></td>
<td></td>
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<tr>
<td>Male</td>
<td>51.54</td>
<td>.009</td>
</tr>
<tr>
<td>Female</td>
<td>66.12</td>
<td>&lt;.001</td>
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Table 3. Factorial model comparison across age groups and genders for the proxy-report version of DISABKIDS-10.

<table>
<thead>
<tr>
<th>Age groups</th>
<th>Model’s goodness of fit</th>
<th>Multi-group analyses</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$\chi^2$(30)</td>
<td>$p$</td>
</tr>
<tr>
<td>Children 8-12</td>
<td>80.77</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Adolescents 13-18</td>
<td>43.78</td>
<td>.05</td>
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<tr>
<td>Gender</td>
<td></td>
<td></td>
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<tr>
<td>Male</td>
<td>71.74</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Female</td>
<td>65.40</td>
<td>&lt;.001</td>
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</table>
1. Do you feel like everyone else even though you have your condition?  .63***
2. Are you free to lead the life you want even though you have your condition?  .54**
3. Is your life ruled by your condition?  -.49**
4. Does your condition bother you when you play or do other things?  -.58**
5. Are you unhappy because of your condition?  -.62**
6. Does your condition get you down?  -.64**
7. Do you feel lonely because of your condition?  -.54**
8. Do you feel different from other children/adolescents?  -.63**
9. Do you think that you can do most things as well as other children/adolescents?  .50**
10. Do your friends enjoy being with you?  .42**
Figure 1. Standardized regression weights of factor loadings for the patient-reported version of the DISABKIDS-10.

Note. e - error.

* $p < .05$; ** $p < .01$, two-tailed.
1. Does your child feel like everyone else even though they have their condition?  

2. Does your child feel free to lead the life they want even though they have their condition?  

3. Does your child feel that their life is ruled by their condition?  

4. Does your child’s condition bother them when they play or do other activities?  

5. Does your child feel unhappy because of his/her condition?  

6. Does your child’s condition get him/her down?  

7. Does your child feel lonely because of his/her condition?  

8. Does your child feel different from other children/adolescents?  

9. Does your child think that he/she can do most things as well as other children?  

10. Does your child feel that their friends enjoy being with them?
Figure 2. Standardized regression weights of factor loadings for the proxy-reported version of the DISABKIDS-10.

Note. e - error.

* \( p < .05 \); ** \( p < .01 \), two-tailed.