Structural validation and dyadic child–parent measurement invariance of the celiac disease quality of life questionnaire

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Objective The celiac disease quality of life questionnaire (CDDUX) is used widely in numerous languages worldwide. However, it's structural and construct validity and child–parent invariance had not been thoroughly examined. The objective of this study was to examine the psychometric properties of the 12-item CDDUX and the extent to which it meets the acceptable requirements of reliability and structural and convergent validity, as well as its child–parent invariance.
Methods In this cross-sectional study, 126 dyads of children aged 8–18 years and their parents completed the Hebrew version self-report and parent-proxy report CDDUX. Recently developed methods to examine psychometric properties and to measure invariance of dyadic samples were used while properly accounting for nonindependence in measurement patterns.
Results A three-factor structure, each with sufficient internal consistency, is confirmed for both children and parents. Removing a single indicator of the diet subscale resulted in full configural ($\chi^2(181) = 202.277$, $P > 0.05$, RMSEA = 0.026) and metric ($\chi^2(189) = 209.543$, $P > 0.05$, RMSEA = 0.043) invariance of the measure between children and parents. However, this occurred only in partial-scalar ($\chi^2(198) = 229.813$, $P > 0.05$, RMSEA = 0.031) and uniqueness invariance, which is nevertheless sufficient for meaningful comparison between the groups.
Conclusion Overall, with minor modifications, the Hebrew version of the CDDUX was found to be a valid measure of children's celiac-related quality of life when measured across children's self-reports and parent-proxy reports. The CDDUX provides meaningful measurement and allows child–parent comparison. Eur J Gastroenterol Hepatol XXX: 00–00

Background

Health-related quality of life (HRQOL) represents a multidimensional subjective perspective in health care that evaluates physical, psychological, social and cognitive components through changing dimensions of time and age [1–3]. Children's HRQOL is an important measure in chronic health conditions, and maintaining good HRQOL is an overall goal of health interventions [4,5]. In the past, determining HRQOL among children with chronic conditions mainly relied on information parents provided concerning their children's physical, rather than social or emotional, aspects [6]. Nowadays, parent-proxy report measures are standard for pediatric psychosocial assessment [7]. Often, both parent- and self-reports are used in research of children. However, children's perspectives differ from those of adults (including their parents), and inconsistencies commonly arise across these reports [8,9].

Celiac disease is a chronic immune-mediated enteropathy caused by ingesting gluten. Maintaining a strict, lifelong gluten-free diet currently is the only available treatment for celiac disease [10]. Outcome measures of HRQOL in celiac disease are complex and multidimensional, and various methodological differences hinder definitive conclusions and produce conflicting results [11]. Diverse HRQOL results have been found among children diagnosed with celiac disease. Some determined no differences compared to healthy controls; others reported reduced quality of life compared to healthy peers but better quality of life compared to children with other chronic conditions [12–14]. Comparing perspectives in children with celiac disease revealed that parents reported lower perceptions of quality of life on parent-proxy reports than did their children on self-reports [15]. Thus, obtaining all HRQOL perspectives is important to view the complete picture of life with celiac disease and help medical professionals understand these perspectives [1,16]. However, comparing parent-reports to their children's HRQOL self-reports can be meaningful only when both the parent and child understand the examined quality of life domain the same way, and neither are substantially influenced by cognitive and motivational biases [17,18]. In other words, when we use the same measure for parents and children, we must ensure both understand each item similarly, and the same items are equally salient in determining the constructs of interests.

One disease-specific pediatric HRQOL tool in research and practice is the celiac disease DUX (CDDUX). The CDDUX is comprised of a self-report and proxy-parent report form [15,19,20]. Despite its increasing use, to our knowledge, no acceptable statistical analysis procedures to

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validate its dimensional structure (i.e., the extent to which the structure of the items and their relatedness to latent factors fit the original and theoretically proposed factorial structure) or its invariance across groups and between children and parents has been published. Therefore, the purpose of this study was to examine the CDDUX psychometric properties and the extent to which it meets acceptable reliability and structural and convergent validity requirements.

Specifically, we first examined the CDDUX structural validity in the Israeli-Hebrew context (i.e., the extent to which its factor structure among children and parents overlaps with the previous and the theoretically motivated three-factor structure [19]) using a confirmatory modeling approach. Next, we examined the CDDUX measurement invariance across self- and parent-reports to establish its convergent validity. We utilized recently developed techniques to model dyadic data and their equivalence across time and groups (dyadic measurement invariance; e.g., [21–25]). These techniques allow us to statistically account for dependencies within dyads while retaining the power of dependent-group data analysis [21].

Methods

Participants

Inclusion criteria were children aged 8–18 years, celiac disease diagnosis confirmed by a physician no less than 6 months before the study, and Hebrew-language proficiency. Children diagnosed with physical or neurological disabilities were excluded. The final sample was 126 children (M = 12.33 years, SD = 2.85) and their parents. The sample included 82 girls and 44 boys, all diagnosed with celiac disease by biopsy (89.7%) or blood tests (10.3%). The majority were diagnosed before the age of 11 years (88.9%), and 67.5% were diagnosed more than 3 years before the study. According to parent reports, 97.6% of the participants 'always' adhered to a gluten-free diet; only three adhered to the diet 'often'. The children studied in grades 2–12 (M = 6.56, SD = 2.95) in mainstream schools. The majority (69%) lived in urban surroundings; the rest lived in the countryside or small communities. The mothers (M = 45 years, SD = 4.79) had an average 16.33 school years (SD = 2.75), and family income was mostly high (63.5%) or average (25.7%).

Procedure

The study was approved by the University of Haifa, Faculty of Social Welfare & Health Sciences Research Ethics Committee (approval number 026/15). The study was performed in accordance with the ethical standards as set forth in the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

Participants were recruited via the Israel Celiac Association, local online celiac disease support groups, and social media. Parents who responded were contacted via email and prescreened for eligibility. Eligible children and parents each signed informed consent forms and completed online questionnaires. Children completed the CDDUX self-report form, and parents completed the proxy form and a demographic and health questionnaire. Parents and children received separate links to the relevant online questionnaires and were instructed to complete the questionnaires independently.

Measures

Demographic and health data for children and parents were collected using an online questionnaire completed by the parent.

Disease-specific HRQOL questionnaire for children with celiac disease [19]. The CDDUX is a self-report HRQOL questionnaire for children with celiac disease aged 8–18 years that provides information about children's perceptions of their condition. The CDDUX is accompanied by a parent-proxy version. Its 12 questions are rated on a 5-point Likert scale with picture questions. Higher scores describe the perception of better HRQOL. The results comprise a total score and three scale scores (1) diet—how the child feels about lifelong dietary adherence, (2) communication—how the child feels when speaking about celiac disease and (3) having celiac disease—the child's feelings when offered or thinking about food containing gluten. In this study, the Hebrew version (translated to and back-translation from Hebrew) was used [26]. The CDDUX authors granted permission for its use in the present study [19].

Data analysis

All analyses were conducted using SPSS AMOS version 21.0 and IBM SPSS Statistics version 25.0. There were no missing data. First, we examined descriptive values, including means, standard deviations, skewness and kurtosis of all items. Second, the initial factor structure of the CDDUX was examined using principal component analysis (PCA) with a maximum-likelihood estimation approach, varimax rotation and Kaiser normalization. We set the item-loadings cut-off value to 0.50 to ensure sufficiently high loadings in later validation stages [27]. Reliabilities of latent factors were obtained using Cronbach's alpha indices.

Third, we conducted multiple confirmatory factor analyses (CFA) using maximum-likelihood estimation with robust standard errors, initially aiming to test two a priori alternative models: a single-factor model and a theory-driven three-factor model. Model fit was improved by allowing a minimal number of item errors to covary, as long as they were on the same factor and the items were similar in phrasing or meaning (as recommended by [28,29]). The most satisfactory model for both children and parents served as a baseline model for equivalence testing.

Reliability was assessed using composite reliability, with composite reliability >0.70 indicating sufficient reliability. Convergent validity was examined using the average variance extracted (AVE), where values above 0.50 and larger than the factor's correlations with other factors confirmed convergence. Discriminant validity was confirmed when the maximum shared variance (MSV) and the average shared squared variance (ASV) were lower than the AVE for all constructs [27].

Fourth, we used recently recommended techniques to test dependent-group invariance in a dyadic structure. This involves modeling observed and latent factors in both groups in a single structure while accounting for the

interrelationship between dyad members by adding covariances between cross-group latent factors and pairs of item errors [22,24]. We followed the recommended invariance-testing sequential order, comparing increasingly constrained models: configural (no equality constrains), metric (equal factor loadings) and scalar invariance (equal intercepts) [30,31]. Although not prerequisite to comparing distributions between groups, uniqueness (residual) invariance also was examined by placing constrains on item residual variances. Failing to achieve full invariance in any stage leads to examining partial invariance, deemed acceptable when at least two loadings or intercepts within each latent factor demonstrates equivalence [32].

In all CFA and measurement invariance tests, the goodness of fit was evaluated following the guidelines in [33]. In addition to the chi-square test as an overall model-fit index, we used two incremental-fit indices: the comparative fit index (CFI) and the Tucker–Lewis index (TLI); two absolute-fit indices—the root-mean-square error of approximation (RMSEA) with 90% confidence interval and the standardized root-mean-square residual (SRMR)—and the Akaike information criterion (AIC) as a predictive-fit indicator. We applied acceptable cut-off points to determine a good model fit (CFI and TLI ≥ 0.95, RMSEA and SRMR ≤ 0.06) [34].

In validating child–parent invariance, nested models were compared using the chi-square difference test. However, because the chi-square test may obscure nonsubstantive discrepancies [35], we also inferred equal fit when the absolute difference in CFI was less than 0.01. Nonnested models were tentatively compared using AIC, with smaller values indicating more accurate models [36].

Finally, if at least partial-scalar invariance was established, then the resulting model served as a baseline to compare latent mean differences by fixing children's (the reference group) factor means to zero and freely estimating them among parents. The significance of the mean difference for each factor was assessed through the composite reliability value, and the magnitude of the difference was evaluated using Cohen's measure of sample effect sizes.

Results

Psychometric properties

Tables 1 and 2 present descriptive parameters for each item among children and parents with factor loadings obtained from the PCA. Skewness of item-score distributions in absolute values ranged between 0.04 and 0.73 among children and between 0.03 and 0.67 among parents. Absolute kurtosis values ranged between 0.09 and 1.02 among children and 0.03 and 0.83 among parents. These values did not indicate any violation from the assumption of normality; therefore, maximum-likelihood estimation could be used further [37].

The varimax rotated PCA solution in each group indicated a three-factor solution overlapping the three CDDUX subscales that explained more than 57% of the variance. The exception was item 4 from the diet subscale, which among children had high loadings on the having celiac disease factor and, among parents, had no sufficiently high loadings on any factor. Consequently, in the CFA, we examined whether removing item 4 improved the factorial structure. Scale reliability indices were high for the diet and *communication* subscales ($\alpha \geq 0.89$) and satisfactory for the having celiac disease subscale (children $\alpha = 0.65$; parents $\alpha = 0.75$).

Confirmatory factor analysis and validity

Fit indices obtained from multiple single-group CFA for children and parents are presented in Table 3. The results indicated that neither M1 (in which all items loaded on a single unidimensional latent factor) nor M2 (in which all CDDUX items loaded on the three theoretically proposed latent factors) fit the data adequately according to the model-fit indices among children or parents. As the exploratory analysis and modification indices indicated, dropping item 4 from the diet subscale resulted in a sufficiently fitting model (M3A). Further inspection of the modification indices showed that a minor respecification, that is, allowing errors for items 11 and 12 (referring to dietary limitations) to correlate (M3B) could significantly

Table 1. Descriptive statistics and principal component analysis results for the children's self-report CDDUX

					Factor loadings[a]		
Item	*M*	*SD*	Skewness	Kurtosis	Diet	Having celiac disease	Communication
1. When I think of food containing gluten, I feel…	55.12	19.33	–0.04	0.18	–	0.43	–
2. When at school I am given food containing gluten, I find it…	44.19	20.38	0.48	–0.17	–	0.72	–
3. Talking about celiac disease with others my age, I find…	73.14	22.52	–0.59	–0.22	–	–	.80
4. Not being able to eat just everything I want, I feel…	42.56	19.78	0.73	0.19	–	0.63	–
5. When someone offers me food that I can't have, I feel…	50.70	20.68	0.39	–0.18	–	.77	–
6. When I have to explain to others what celiac disease is, I feel…	71.40	21.31	–0.54	–0.09	–	–	0.81
7. Talking about celiac disease, I find…	72.33	21.23	–0.49	–0.13	–	–	0.81
8. Having to follow a lifelong diet, I find…	60.47	25.95	–0.09	–1.02	0.71	–	–
9. Having to pay attention to what I eat, I find…	63.72	21.73	–0.18	–0.38	0.62	–	–
10. Having celiac disease is…	59.30	25.12	–0.11	–0.90	0.75	–	–
11. Not being able to eat all the things other people eat, I find…	46.28	21.14	–0.67	–0.03	0.58	0.50	–
12. Following a diet for my celiac disease is…	59.42	22.73	–0.09	–0.61	0.76	–	–
Eigenvalue	–	–	–	–	2.89	2.28	2.37
% Variance explained	–	–	–	–	24.09	19.03	19.73
Cronbach's alpha[b]	–	–	–	–	0.89	0.75	0.90

N = 172. Factor loadings of 0.40 and above are presented; range: 20–100; cumulative variance explained (%) = 62.86; Kaiser–Meyer–Olkin test = 0.90.
CDDUX, celiac disease quality of life questionnaire.
[a]Rotated factor solution (maximum likelihood with varimax rotation).
[b]Item 4 was excluded from the diet subscale.

Table 2. Descriptive statistics and principal component analysis results for the parent-proxy CDDUX

Item	*M*	*SD*	Skewness	Kurtosis	Factor loadings[a]		
					Diet	Having celiac disease	Communication
1. When my child thinks of food containing gluten, he/she feel…	49.07	16.66	0.03	0.37	–	0.52	–
2. When at school my child is given food containing gluten, he/she finds it…	42.44	18.25	0.64	0.37	–	0.65	–
3. Talking about celiac disease with others his/her age, my child finds…	71.16	19.13	–0.27	–0.17	–	–	0.71
4. Not being able to eat just everything he/she wants, my child feels…	40.70	17.02	0.45	–0.46	–	0.40	–
5. When someone offers my child food that he/she can't have, he/she feels…	45.47	15.91	0.03	–0.20	–	0.61	–
6. When my child has to explain to others what celiac disease is, he/she feels…	71.51	17.30	–0.26	–0.05	–	–	0.94
7. Talking about celiac disease my child finds…	70.12	19.41	–0.54	0.20	–	–	0.85
8. Having to follow a lifelong diet, my child finds…	52.44	19.97	0.10	–0.57	0.75	–	–
9. Having to pay attention to what he/she eats, my child finds…	60.23	19.34	–0.06	–0.03	0.72	–	–
10. Having celiac disease is…	49.42	19.52	0.35	–0.30	0.72	–	–
11. Not being able to eat all the things other people eat, my child finds…	43.84	16.38	0.67	0.83	0.64	–	–
12. Following a diet for his/her celiac disease is…	56.16	19.02	0.11	–0.16	0.75	–	–
Eigenvalue	–	–	–	–	2.88	1.62	2.38
% variance explained	–	–	–	–	23.98	13.47	19.79
Cronbach alpha[b]	–	–	–	–	0.89	0.65	0.89

N = 172. Factor loadings of 0.40 and above are presented; range: 20–100; cumulative variance explained (%) = 57.24.86; Kaiser–Meyer–Olkin test = 0.86.
CDDUX, celiac disease quality of life questionnaire.
[a]Rotated factor solution (maximum likelihood with varimax rotation).
[b]Item 4 was excluded from the diet subscale.

Table 3. Single-group confirmatory factor analysis for children and parents CDDUX with model fit comparisons

Model	χ^2	*df*	CFI	TLI	RMSEA (90% CI)	SRMR	AIC
Children's CDDUX							
M1 one factor (unidimensional)	340.573***	54	0.749	0.693	0.176 (0.159, 0.194)	0.103	388.573
M2 CDDUX theoretical (three factors)	108.862**	51	0.949	0.934	0.081 (0.060, 0.303)	0.063	162.862
M3A CDDUX revised (exc. item 4)	56.225	41	0.985	0.980	0.047 (0.000, 0.075)	0.048	106.225
M3B CDDUX revised and modified	49.232	40	0.991	0.988	0.037 (0.000, 0.068)	0.046	101.232
Parents' CDDUX							
M1 one factor (unidimensional)	367.353***	54	0.676	0.605	0.184 (0.167, 0.277)	0.120	415.353
M2 CDDUX theoretical (three factors)	85.452**	51	0.964	0.954	0.063 (0.038, 0.086)	0.057	139.452
M3A CDDUX revised (exc. item 4)	60.736*	41	0.979	0.971	0.053 (0.020, 0.080)	0.047	110.736
M3B CDDUX revised and modified	45.544	40	0.994	0.992	0.028 (0.000, 0.062)	0.046	97.544

CDDUX revised = excluding item 4; CDDUX revised and modified = excluding item 4, including covariance between errors of items 11 and 12.
AIC, Akaike information criterion; CDDUX, celiac disease quality of life questionnaire; CFI, comparative fit index; CI, confidence interval; RMSEA, root-mean-square error of approximation; SRMR, standardized root-mean-square residual; TLI, Tucker–Lewis index.
*$P < 0.05$.
**$P < 0.01$.
***$P < 0.001$.

Table 4. Convergent and discriminant validity assessment in the revised and modified CDDUX model among children and parents

Latent construct	Composite reliability	AVE	MSV	ASV
Children's CDDUX				
Having celiac disease	0.756	0.512	0.460	0.336
Diet	0.897	0.636	0.460	0.426
Communication	0.899	0.747	0.393	0.302
Parents' CDDUX				
Having celiac disease	0.653	0.388	0.408	0.236
Diet	0.895	0.631	0.408	0.303
Communication	0.896	0.744	0.199	0.131

Model specifications are for the revised and modified model (excluding item 4, including a covariate between items 11 and 12).
ASV, average shared squared variance; AVE, average variance extracted; CDDUX, celiac disease quality of life questionnaire; MSV, maximum shared variance.

improve the model fit among both children ($r = 0.60$, $P < 0.001$, model comparison: $\Delta\chi^2(1) = 6.993$, $P = 0.008$, ΔCFI = 0.006) and parents ($r = 0.53$, $P < 0.001$, model comparison: $\Delta\chi^2(1) = 15.192$, $P < 0.001$, ΔCFI = 0.015). Consequently, M3B was used as a baseline model in the following dyadic measurement invariance tests.

Results of the validity assessment (Table 4) showed sufficiently high reliabilities and AVE for all factors, except for having celiac disease among parents, which demonstrated slightly lower composite reliability and AVE values than required (>0.70 and >0.50, respectively). Moreover, for this factor, the AVE was slightly smaller than the MSV, pointing out possible discriminant validity issues. The composite reliability and AVE values, in this case, were not improved to the typically acceptable threshold when removing observed items [38].

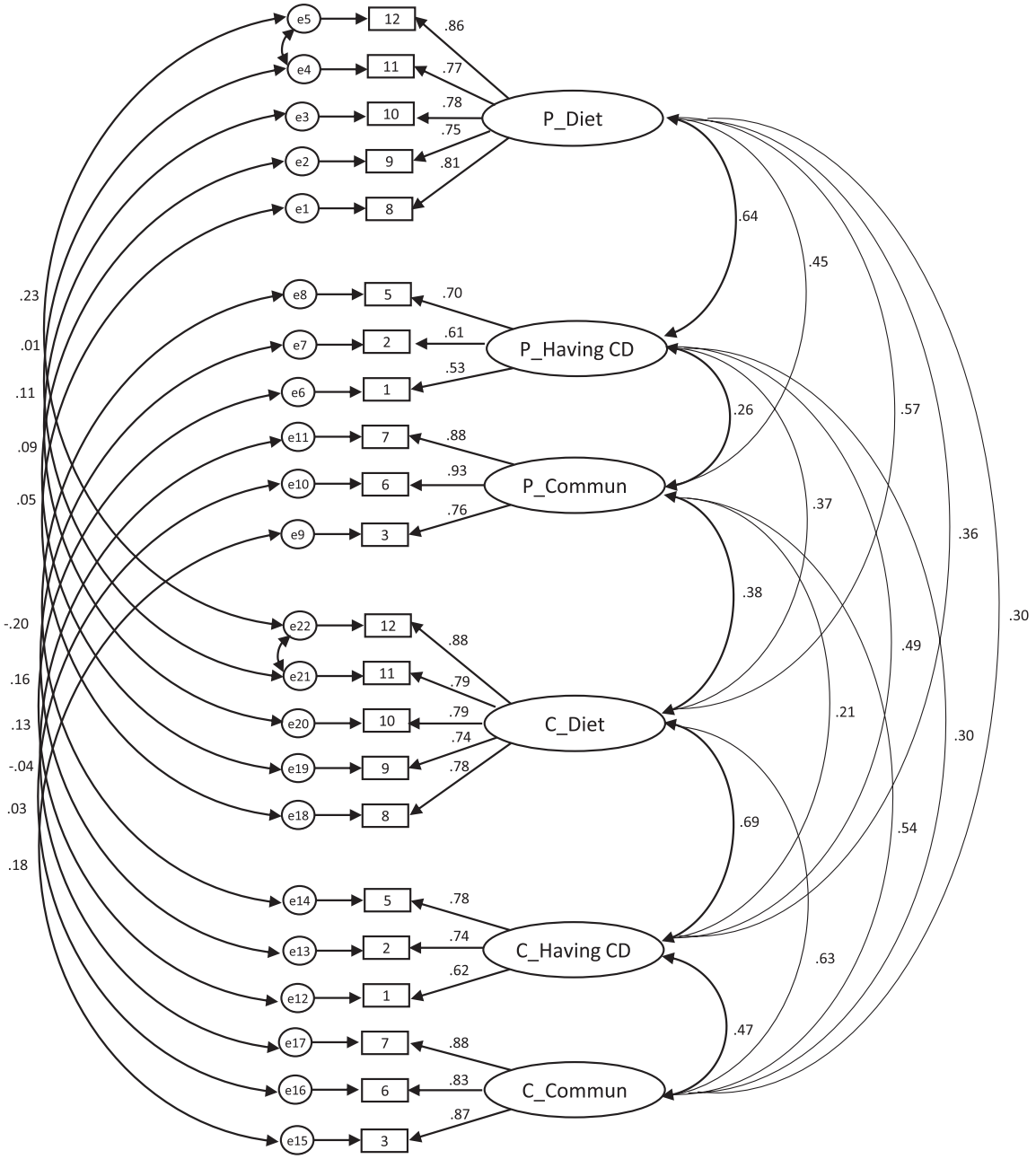

Fig. 1. Full dependent-group configural invariance model for children and parent CDDUX. $N = 172$. $\chi^2(df = 2) = 202.277$, $P = 0.133$, RSMEA = 0.026, CFI = 0.990. Latent factors (circles) beginning with P belong to parents and beginning with C to children. Observed variables (rectangles) numbers represent CDDUX item numbers. Coefficients are standardized. The model includes covariances between pairs of item errors (left side) and between all latent variables (right side) in order to account for nonindependence in the data. CDDUX, celiac disease quality of life questionnaire.

Accounting for nonindependence and measurement invariance

A configural-invariance model for child–parent dyads was specified and estimated. The model used the previously obtained baseline model (M3B) and included 22 observed variables (i.e., 11 items per group) and six latent factors (representing the three subscales in each group).

To determine the extent to which we should account for nonindependence, we first examined bivariate correlations between pairs of identical items for children and parents. All correlations were significant ($P < 0.05$) and ranged between $r = 0.18$ for item 5 and $r = 0.52$ for item 12. Already, this supported the need to account for interdependencies in the invariance testing. We then tested whether including freely estimated correlated error terms between common items for children and parents (e.g., correlating the error term of item 1 for children with the error term of item 1 for parents), as well as covariances between all cross-group latent factors, improved the model fit. We found that including these interrelationships significantly improved the fit of the configural model (comparison indices: $\Delta\chi^2(20) = 130.752$, $P < 0.001$, ΔCFI = 0.053). Hence, we retained these covariance and uniqueness interdependencies in subsequent invariance analyses.

The configural model (MI1, depicted in Fig. 1) showed an excellent model fit according to all indices (Table 5). Due to small deviation from multivariate normality (multivariate kurtosis = 61.62, $z = 12.43$), we also examined

Table 5. Parent-child dyadic measurement invariance tests for the Hebrew-version CDDUX

Invariance model	χ^2	*df*	CFI	TLI	RMSEA (90% CI)	SRMR	AIC	Comparisons between nested models.	$\Delta\chi^2$ (Δdf)	*P* value	ΔCFI	Invariant?
MI1 configural	202.277	181	0.990	0.987	0.026 (0.000, 0.044(	0.047	390.277	–	–	–	–	Full
MI2 metric	209.543	189	0.990	0.988	0.025 (0.000, 0.043)	0.046	381.543	M1-M2	7.266 (8)	0.508	0.000	Full
MI3a full-scalar	256.421	200	0.973	0.969	0.041 (0.024, 0.055)	0.046	406.421	M3a-M2	46.878	<0.001	0.019	No
MI3b partial-scalar (items 8 and 10 not constrained)	229.813	198	0.985	0.985	0.031 (0.000, 0.047)	0.046	383.813	M3b-M2	20.270	0.016	0.005	Partial
MI4a full-uniqueness, partial-scalar	267.798	207	0.971	0.971	0.041 (0.025, 0.055)	0.048	403.798	M4a-M3b	37.986	<0.001	0.014	No
MI4b partial-uniqueness (items 6 and 11 not constrained)	236.689	205	0.985	0.985	0.030 (0.000, 0.046)	0.047	376.689	M4b-M3b	6.877	0.442	0.003	Partial

AIC = Akaike information criterion; CDDUX, celiac disease quality of life questionnaire; CI, confidence interval.

Table 6. Comparison between paired child- and parent-reported CDDUX subscales and intergroup correlations

	Children		Parents				
CDDUX subscales	*M*	*SD*	*M*	*SD*	*t*(171)	*P* value	Child–parent correlation (*r*)
Diet[a]	56.473	18.807	53.411	15.564	2.33	0.02	0.51*
Having celiac disease	50.078	16.401	45.659	13.038	3.44	0.001	0.36*
Communication	72.287	19.776	70.930	16.859	–0.96	0.337	0.50*

Scores are unweighted averages converted to 20–100 to facilitate comparison with previous studies.
CDDUX, celiac disease quality of life questionnaire.
[a]Excluding items 4, 8 and 10.
*$P < 0.001$.

the Bollen–Stine bootstrap *P* value, which at $P = 0.58$ further confirmed the model was correct. According to a chi-square difference test and CFI change between MI1 and MI2, full-metric invariance also was achieved, suggesting that cross-group factor loadings were proportionally equivalent. However, the full-scalar invariance (MI3a) was not supported (the chi-square change from MI2 was significant, $P < 0.001$, and the change in CFI was larger than 0.01). Consequently, partial invariance was attempted by examining modification indices, which suggested unequal intercepts for items 8 and 10 (diet subscale). In both items, the intercept was higher among children (difference of 0.401 and 0.494, respectively). Relaxing the intercepts of these two items resulted in sufficient invariance on the scalar level (MI3b).

In the test for uniqueness invariance (MI4a), only residual variances for items found to be scalarly invariant were constrained to be equal between children and parents (that is, the error terms of items 8 and 10 were freely estimated in both groups). We compared each item's residual variance across children and parents and found the variance was larger for children than for parents on item 6 (difference = 0.236) and item 11 (difference = 0.168). Relaxing the constrains on the residuals of these two items (MI4b) resulted in statistically insignificant chi-square and CFI differences (compared to MI4a), supporting partial-uniqueness invariance of the CDDUX between children and parents. Furthermore, MI4b demonstrated the best fit according to fit indices, which strengthened the overall partial and sufficient invariance of the revised Hebrew-version CDDUX between parents and children on all acceptable measurement invariance levels.

Structural invariance

Finally, the partial-scalar invariance model (MI3b) was used to compare latent means between children and parents (Table 6). The results indicated that parents had higher mean scores on diet (estimated mean difference: –0.165, $SE = 0.072$, composite reliability = –2.297, $P = 0.022$) and on having celiac disease (estimated mean difference: –0.237, $SE = 0.072$, composite reliability = –3.274, $P = 0.001$), with rather small effect sizes ($d = 0.18$ and $d = 0.34$, respectively). No differences were found for communication (estimated mean difference: –0.067, $SE = 0.074$, composite reliability = –0.905, $P = 0.365$). These findings are similar to those obtained by paired *t* tests for unweighted average scores across the CDDUX subscales.

Discussion

The CDDUX often is used to assess the quality of life of children with celiac disease in a dyadic nature, considering both the child's self-report and the parent-proxy report of its constructs. In this paper, we demonstrated how the assumption that children and parents similarly understood and conceptualized the CDDUX HRQOL statements, which stands behind conventional comparisons of scores between children's and parents' ratings, can be estimated statistically through measurement invariance or equivalence tests. Accordingly, it is necessary to measure the dyadic to avoid biased parameter estimates and errors [24]. Establishing equivalence of the CDDUX implies absent or minimal cross-generation biases in measurements and that the psychometric properties though which the observed and latent variables relate to each other are similar between children and parents.

In the group-specific analysis, the CDDUX demonstrated satisfying indices for convergent and discriminant validity across the three subscales, with two exceptions. First, item 4 in the diet subscale had cross- (children) or low (parents) factor loading, and its inclusion in the

confirmatory models resulted in unsatisfying fit indices. Thus, we excluded this item from the core of our analyses. When examining the content and context of item 4 ('Not being able to eat just everything I want . . .'), it seems that of all the CDDUX items, this statement conferred the least celiac disease–specific meaning. Possibly, this influenced its suitability to the models.

Second, indicators for having celiac disease among parents fell below the acceptable threshold for construct validity in a confirmatory analysis. This can indicate that its items do not explain well the latent factor of having celiac disease. Interestingly, previous reports of reliability indices also showed lower consistency of items for having celiac disease compared to the other subscales [19,39]. Possibly, this shortcoming reflects the concept of self-reporting one's chronic health condition and its subjective meaning to each individual [3]. Although parents are considered reliable informants in pediatric assessments [7], they nevertheless may find it difficult to assess their child's personal feelings on having celiac disease, as opposed to feelings related to talking about the disease (communication) or to the restricted diet (diet), which may be more visible to parents managing their child's condition.

From a statistical perspective, it has been suggested that AVE can be lower than 0.50 when composite reliability values are higher than 0.60 [40]. This held in all constructs in the CDDUX model for children and parents. Moreover, all factor loadings of *having celiac disease* items for parents were above 0.50 and significant, and its associated AVE (0.388) was not much higher than the AVS value (0.236), suggesting that convergent and discriminant validity can be established. Considering the convention that AVE values slightly below 0.50 can be disregarded somewhat in exploratory and initial studies [38], these results emphasize the need to further investigate the CDDUX psychometric properties in other contexts and its equivalence both between parents and children and across socio-cultural contexts and languages.

The test of child–parent invariance satisfied the need for full configural and metric equivalence, suggesting that the CDDUX conceptual framework is the same for children and parents and that its items are roughly equally salient for children and parents within subscales. That is, children and parents share an understanding of the three subscales of the CDDUX.

However, on the scalar level, only partial analysis was established. Failing to achieve full scalar-level invariance suggests that children and parents do not fully understand and conceptualize the CDDUX similarly. Specifically, the analysis indicated that two diet subscale items (items 8 and 10) had invariant intercepts, and these items' means were higher in children than in parents. Both item 8 ('Having to follow a lifelong diet . . .') and item 10 ('Having celiac disease is . . .') refer to long-term implications of celiac disease. Perhaps the gap in perspectives specifically in these items reflects the children's and parents' different cognitive-development levels. The age range of participants in this study includes the transition stage from childhood to adolescence, a time when adolescents become more interested in the future, and the cognitive ability to make decisions while considering the future increases [41]. Consequently, we cannot rule out that perceptive differences between children and parents or measurement biases affect the factor means when these two items are included as observed indicators of the diet latent factor. Therefore, we excluded them from our analysis of differences in mean scores. Nevertheless, it is important to stress that nine items (three from each subscale) demonstrated acceptable invariance at the configural, metric and scalar levels, prompting meaningful mean comparison according to acceptable practices [32]. Accordingly, based on our analysis, we conclude that an alternative but sufficiently valid nine-item measure of the CDDUX can achieve full-scalar invariance and be used as a highly valid tool to both assess the HRQOL of children with celiac disease and substantively compare child and parent ratings.

Moreover, only partial-uniqueness invariance was achieved, with unequal residuals for items 6 and 11 from the communication and the diet subscales, respectively. The larger residual variances for children compared to their parents suggests these items might have higher unexplained errors among children or are not as strong indicators of the child's quality of life in terms of diet and communication for children as they are for parents. Regardless, it is important to emphasize that strict forms of invariance are very difficult to obtain when measuring psychological constructs [31,32]. Furthermore, invariance at this level is required only when specific item scores are compared across groups [30,32]. Because such high invariance levels were tenable for seven CDDUX items, specific item comparison is indeed possible for the majority of items and across the three CDDUX subscales.

Achieving partial-scalar invariance enabled us to examine structural means and compare the mean level of each latent factor between children and parents. Parents, on average, evaluated their children's quality of life more negatively than did the children themselves on the diet and having celiac disease subscales. These results correspond to findings that parents of children with chronic health conditions tend to report lower HRQOL than do their children [42,43]. Moreover, the results align with findings of the CDDUX across cultures and languages, specifying lower parent-reported HRQOL [15]. However, these effects were rather small in magnitude.

Although differences between latent means were small, group scores were not interchangeable between child and parent reports, as indicated by low-to-moderate same-factor correlations. Evidently, parents' scores explain no more than 25% of their children's scores on the CDDUX. This, in a sense, rules out the possibility to substitute child self-reports with parent-proxy reports.

Limitations and future directions

Due to the cross-sectional design, the resultant convenience sample was composed largely of participants who are members of the Israel Celiac Association or active on various social media platforms. Thus, the results do not necessarily represent all Israeli–Hebrew speaking children with celiac disease and their parents in Israel. The results of our measurement invariance test and the suggestion to drop or modify specific items can be valid only for the specific socio-cultural context in which the study was conducted (Hebrew speakers in Israel) and are not necessarily valid for other cultural settings. The extent to which we should prompt researchers and practitioners to

drop noninvariant items when comparing children's and parents' ratings should be conditioned on future studies that examine the CDDUX invariance not only within its primary dyadic structure of children and their parents but also in larger samples across different groups, languages and countries (i.e., cross-cultural invariance) and desirably, across time (i.e., longitudinal invariance).

Moreover, although an important advantage of the CDDUX is its short length, additional dimensions of HRQOL, such as future orientation of life with celiac disease, especially among adolescents who are in the midst of the critical developmental stage toward adulthood.

Conclusion

To our knowledge, this is the first attempt to validate the CDDUX factorial structure and establish its equivalence across children and parents using confirmatory methods. Moreover, our novel approach to invariance testing considered the detected interdependencies between children and parents by modeling dyads—rather than individual respondents—as the analysis unit. This approach enabled more accurate invariance estimation than would the more conventional multigroup method [21,24]. Overall, with minor modifications, the Hebrew version of the CDDUX was found to be a valid measure of children's HRQOL when measured across children's self-reports and parents' proxy reports, allowing meaningful comparison between the distributions. Further examinations of the CDDUX measurement invariance can be useful for modifying the CDDUX to obtain a highly valid measure that allows reaching child–parent agreement and additional intergroup comparisons. Therefore, the CDDUX questionnaire can been used as a valid tool by health care providers to gain a better understanding of the children's perceptions and to adopt suitable interventions to improve their HRQOL when needed.

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Conflicts of interest

There are no conflicts of interest.

References

1 Eisen GM, Locke GR 3rd, Provenzale D. Health-related quality of life: a primer for gastroenterologists. *Am J Gastroenterol* 1999; 94:2017–2021.
2 Renwick R. Quality of life for young people with neurological and developmental conditions: issues and challenges. In: Ronen G, Rosenbaum P, editors. *Life Quality Outcomes in Children, Young People with Neurological, Developmental, Conditions*. London: Mac Keith Press; 2013. pp. 22–35.
3 de Wit M, Hajos T. Health-related quality of life. In: Gellman MD, Turner JR, editors. *Encyclopedia of Behavioral Medicine*. New York: Springer; 2013. pp. 929–931.
4 Hanson CL. Quality of life in families of youths with chronic conditions. In: Koot J, Wallander JL, editors. *Quality of Life in Child and Adolescent Illness: Concepts, Methods and Findings*. London: Routledge; 2001. pp. 181–209.
5 Guyatt GH, Feeny DH, Patrick DL. Measuring health-related quality of life. *Ann Intern Med* 1993; 118:622–629.
6 Eiser C, Varni JW. Health-related quality of life and symptom reporting: similarities and differences between children and their parents. *Eur J Pediatr* 2013; 172:1299–1304.
7 Cushman GK, Stolz MG, Shih S, Blount R, Otley A, Talmadge C, *et al*. Parent IMPACT-III: development and validation of an inflammatory bowel disease-specific health-related quality-of-life measure. *J Pediatr Gastroenterol Nutr* 2020; 70:205–210.
8 de Los Reyes A. Introduction to the special section: more than measurement error; discovering meaning behind informant discrepancies in clinical assessments of children and adolescents. *J Clin Child Adolesc Psycho* 2011; 40:1–9.
9 Fattore T, Mason J, Watson E. Activities as autonomy and competence: the meaning and experience of leisure for well-being. In: Fattore T, Mason J, Watson E, editors. *Children's Understandings of Well-Being*. vol. 14. The Netherlands: Springer; 2017. pp. 147–192.
10 Lindfors K, Ciacci C, Kurppa K, Lundin KEA, Makharia GK, Mearin ML, *et al*. Coeliac disease. *Nat Rev Dis Primers* 2019; 5:3.
11 Ludvigsson JF, Ciacci C, Green PH, Kaukinen K, Korponay-Szabo IR, Kurppa K, *et al*. Outcome measures in coeliac disease trials: the Tampere recommendations. *Gut* 2018; 67:1410–1424.
12 de Lorenzo CM, Xikota JC, Wayhs MC, Nassar SM, de Souza Pires MM. Evaluation of the quality of life of children with celiac disease and their parents: a case-control study. *Qual Life Res* 2012; 21:77–85.
13 Howard RA, Urquhart-Law G. Psychological well-being of children and young people with coeliac disease. In: Martin C, Dovey T, editors. *Paediatric Gastrointestinal Disorders*. London: CRC Press; 2019. doi: 10.1201/9780429091346-12.
14 Rashid M, Cranney A, Zarkadas M, Graham ID, Switzer C, Case S, *et al*. Celiac disease: evaluation of the diagnosis and dietary compliance in Canadian children. *Pediatrics* 2005; 116:e754–e759.
15 Nikniaz Z, Abbasalizad Farhangi M, Nikniaz L. Systematic review with meta-analysis of the health-related quality of life in children with celiac disease. *J Pediatr Gastroenterol Nutr* 2020; 70:468–477.
16 Rojas M, Oyarzún A, Ayala J, Araya M. Health related quality of life in celiac children and adolescents. *Rev Chil Pediatr* 2019; 90:632–641.
17 Borsboom D. When does measurement invariance matter? *Med Care* 2006; 44:S176–S181.
18 Funder DC. On seeing ourselves as others see us: self-other agreement and discrepancy in personality ratings. *J Pers* 1980; 48:473–493.
19 van Doorn RK, Winkler LM, Zwinderman KH, Mearin ML, Koopman HM. CDDUX: a disease-specific health-related quality-of-life questionnaire for children with celiac disease. *J Pediatr Gastroenterol Nutr* 2008; 47:147–152.
20 Weiner NW, Toglia J, Berg C. Weekly calendar planning activity (WCPA): a performance-based assessment of executive function piloted with at-risk adolescents. *Am J Occup Ther* 2012; 66:699–708.
21 Claxton SE, Deluca HK, Van Dulmen MHM. Testing psychometric properties in dyadic data using confirmatory factor analysis: current practices and recommendations. *Test Psychom Methodol Appl Psychol* 2015; 22:181–198.
22 Sakaluk JK, Kilshaw RE, Fisher AN, Leshner CE. Dyadic measurement invariance and its importance for replicability in romantic relationship research. PsyArXiv Preprints 2019. https://doi.org/10.31234/osf.io/9vcnz
23 Bluemke M, Jong J, Grevenstein D, Mikloušić I, Halberstadt J. Measuring cross-cultural supernatural beliefs with self- and peer-reports. *PLoS One* 2016; 11:e0164291.
24 Kenny DA, Kashy DA, Cook WL. *Dyadic Data Analysis*. New York: Guilford Press; 2006.
25 Ledermann T, Kenny DA. Analyzing dyadic data with multilevel modeling versus structural equation modeling: a tale of two methods. *J Fam Psychol* 2017; 31:442–452.
26 Meyer S, Rosenblum S. Children with celiac disease: health-related quality of life and leisure participation. *Am J Occup Ther* 2016; 70:7006220010p1–7006220010p8.
27 Hair JF, Anderson RE, Babin BJ, Black WC. *Multivariate Data Analysis: a Global Perspective*. Vol. 7. London: Pearson; 2010.
28 Brown TA. *Confirmatory Factor Analysis for Applied Research*. New York: Guilford Press; 2015.
29 Hermida R. The problem of allowing correlated errors in structural equation modeling: concerns and considerations. *Comput Methods Soc Sci* 2015; 3:5–17.

30 Milfont TL, Fischer R. Testing measurement invariance across groups: applications in cross-cultural research. *Int Psycho Stud* 2010; 3:111–130.
31 de Schoot R, Lugtig P, Hox J. A checklist for testing measurement invariance. *Eur J Dev Psychol* 2012; 9:486–492.
32 Byrne BM, Shavelson RJ, Muthén B. Testing for the equivalence of factor covariance and mean structures: the issue of partial measurement invariance. *Psychol Bull* 1989; 105:456–466.
33 Worthington RL, Whittaker TA. Scale development research: a content analysis and recommendations for best practices. *Couns Psychol* 2006; 34:806–838.
34 Hu L, Bentler PM. Cutoff criteria for fit indexes in covariance structure analysis: conventional criteria versus new alternatives. *Struct Equ Modeling* 1999; 6:1–55.
35 Cheung GW, Rensvold RB. Evaluating goodness-of-fit indexes for testing measurement invariance. *Struct Equ Modeling* 2002; 9:233–255.
36 West SG, Taylor AB, Wu W. Model fit and model selection in structural equation modeling. In Hoyle RH, editor. *Handbook of Structural Equation Modeling*. New York: Guilford Press; 2012. pp. 209–231.
37 Finney S, DiStefano C. Non-normal and categorical data in structural equation modeling. In Hancock GR, Mueller RO, editors. *Structural Equation Modeling: a Second Course.* Greenwich: Information Age; 2006. pp. 269–314.
38 Ping RA. Is there any way to improve average variance extracted (AVE) in a latent variable (LV) X (Revised)? 2009. Retrieved December 22, 2020, from http://webcache.googleusercontent.com/search?q=-cache:IAgwWhv6W8wJ:www.wright.edu/~robert.ping/ImprovAVE2.doc+&cd=1&hl=de&ct=clnk&gl=de.
39 Lins MT, Tassitano RM, Brandt KG, Antunes MM, Silva GA. Translation, cultural adaptation, and validation of the celiac disease DUX (CDDUX). *J Pediatr (Rio J)* 2015; 91:448–454.
40 Fornell C, Larcker DF. Evaluating structural equation models with unobservable variables and measurement error. *J Mark Res* 1981; 18:39–50.
41 Vroman K. Adolescent development: transitioning from child to adult. In: Case-Smith J, Clifford O'Brien J, editors. *Occupational Therapy for Children and Adolescents*. 7th ed. St. Louis: Elsevier; 2015. pp.102–121.
42 Ingerski LM, Modi AC, Hood KK, Pai AL, Zeller M, Piazza-Waggoner C, *et al*. Health-related quality of life across pediatric chronic conditions. *J Pediatr* 2010; 156:639–644.
43 Upton P, Lawford J, Eiser C. Parent-child agreement across child health-related quality of life instruments: a review of the literature. *Qual Life Res* 2008; 17:895–913.