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Cross-Diagnostic Validity of the Congenital Glaucoma Caregiver's Quality of Life Questionnaire (CarCGQoL)

Vijaya K. Gothwal[1], Sujata Sharma[1], and Anil K. Mandal[2]

[1] Brien Holden Eye Research Centre- Patient Reported Outcomes Unit, L V Prasad Eye Institute, Hyderabad, India
[2] Jasti V Ramanamma Children's Eye Care Centre, L V Prasad Eye Institute, Hyderabad, India

Correspondence: Vijaya K. Gothwal, Brien Holden Eye Research Centre, Patient-Reported Outcomes Unit, L V Prasad Eye Institute, Kallam Anji Reddy Campus, LV Prasad Marg, Banjara Hills, Hyderabad, 500034, Telangana, India. e-mail: vijayagothwal@gmail.com

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Purpose: The Caregiver's Congenital Glaucoma QoL (CarCGQoL) questionnaire was proposed as a measure of QoL of caregivers with children with primary congenital glaucoma (PCG). Support for its psychometric properties among other diagnostic groups is required for scores to be interpreted in the same manner across groups. Therefore we investigated the measurement properties and cross-diagnostic validity of the CarCGQoL questionnaire among caregivers of children with congenital cataract, retinopathy of prematurity (ROP), and blinding corneal disorders.

Methods: Eight hundred ninety-one caregivers (mean age, 28.3 years; 76% mothers) of children with congenital cataract (n = 407), ROP (n = 272), and blinding corneal disorders (n = 212) completed the CarCGQoL questionnaire. Rasch analysis was used to investigate the psychometric properties. Unidimensionality (by principal components analysis of residuals, PCA) was examined for each group and for pooled sample. Differential item functioning (DIF) was investigated to explore whether bias in responses to the questionnaire existed for certain subgroups as compared to the reference group (PCG).

Results: Across groups, six items necessitated removal because of misfit (two common and four uncommon), after which three different versions of the questionnaire emerged. Measurement precision was adequate for each group and for the pooled sample (0.80). Unidimensionality was observed, albeit with some DIF. Regardless of the level of QoL, caregivers in the pooled sample were more likely to endorse two items reflecting ability to face child's disease, and interest to pursue leisure activity, as compared with caregivers of children with PCG.

Conclusions: Care must be taken when data from the CarCGQoL questionnaire from different pediatric ocular conditions are pooled, given the presence of DIF between the reference group (PCG) and the pooled sample.

Translational Relevance: When evaluating the impact of interventions on the caregiver's QoL using the CarCGQoL questionnaire in a pooled sample of pediatric ocular conditions, cross-diagnostic DIF must be taken into account.

Introduction

Caring for children with chronic pediatric ocular conditions such as retinoblastoma, congenital glaucoma, corneal disorders, and more is often challenging and affects family life.[1,2] Meeting the high care demands of affected children requires much time, effort, and patience, resulting in psychological distress, depression, anxiety, and other mental or physical health problems among the caregivers.[3,4] Caregivers may experience fatigue, bodily pain, or sleep disruption given the increased stress arising from the need for constant vigilance of the affected child.[5–7] Many caregivers also suffer financially, given high out-of-pocket health care expenses, loss of wages, underemployment, or loss of employment.[3] Some caregivers with extreme stress are at a higher risk of marital breakdown or failing to provide adequate care for themselves and their child and are unable to work given the singular focus on the affected child that takes precedence over others' needs and wishes in

the family.[8,9] Taken together, it can be surmised that the child's disease can have an adverse impact on the caregiver's quality of life (QoL).[10]

Children with chronic ocular conditions require long-term care with their treating ophthalmologist and may require change in the management plan in terms of additional surgeries, medications, and replacement of glasses or other equipment during the course of the follow-up. The events associated with prolonged care can adversely impact the caregiver's QoL, so it is important to measure the impact that the demands of caregiving may have on the lives of the parents of these children. It is essential that the caregiver's QoL is preserved because a healthy emotional well-being of parents could in turn positively influence the effect of any interventions performed in children with chronic ocular conditions. Moreover, tracking the progress of caregiver's QoL in children with chronic ocular pediatric conditions can be a useful marker to gauge the disadvantages and benefits of treatment provided to the child. If an intervention improves the treatment outcomes of a given pediatric ocular condition, it will likely not only improve the child's visual status but also help improve the caregiver's QoL.[2] Such a QoL instrument can also serve as a useful outcome measure for interventions aimed directly at caregivers to document the impact of supportive care over time.[11]

Although it has been argued that all chronic illnesses can negatively impact the QoL of the parents of children with chronic illness, little is known about the caregiver's QoL among children with chronic ocular conditions. This lacuna may partly be attributed to the unavailability of suitable instruments for assessment. In an effort to bridge this gap in the literature, we developed the Caregiver Congenital Glaucoma QoL Questionnaire (CarCGQoL) using Rasch analysis in caregivers of children with primary congenital glaucoma (PCG) and demonstrated it to have robust psychometric properties.[1] The CarCGQoL questionnaire provides a unidimensional measure of QoL among caregivers of children with PCG. Given that the birth of a child with an eye defect adversely affects the QoL of the caregivers,[12] it can be expected that the QoL deficits in caregivers of children with PCG are similar to caregivers of children with other chronic ocular conditions, hence, warranting the validation of the CarCGQoL questionnaire in these groups..

Ideally, a questionnaire should only be used for the specific condition(s) for which it has been developed and tested. However, there are several examples in health care where questionnaires, while originally developed for generic use or for some specific conditions, have been used in other conditions. Some of the examples include the Short-Form 36 (SF-36; developed for healthy persons and patients), Visual Function-14 (VF-14; developed for cataract), and Impact of Vision Impairment Profile (IVI; developed for visually impaired), which have been demonstrated to be valid in other disease groups too.[13–16] This is because these questionnaires contain some items that are relevant and applicable across disease groups. However, it is important that the questionnaires fulfil several psychometric requirements in different disease groups before they can be used confidently.

Thus the overall aim of this study was to validate the CarCGQoL questionnaire when used in samples of caregivers of children with congenital cataract, ROP, and blinding corneal conditions to investigate whether the questionnaire could be used as a generic scale. In addition, we examined item bias by comparing item functioning in caregivers of PCG (our previous questionnaire development study) and the pooled sample of caregivers of children from the present study. Evaluation of the CarCGQoL questionnaire using Rasch analysis with a mixed sample would provide information on psychometric properties on a more generic level and whether the scores work the same way and have the same meaning in different patient groups.

Methods

Participants

We recruited a heterogeneous sample consisting of parents (caregivers) of children with congenital cataract (n = 407), ROP (n = 272), and blinding corneal disorders (e.g., congenital hereditary endothelial dystrophy; n = 212) scheduled to undergo surgery (treatment naive) at a tertiary eye care center in South India. All the parents were 18 years or older. One caregiver for each child was recruited, defined as a person living with the child and looking after the child for most part of the time, and being the primary caregiver for the patient. Eligible participants spoke one of the three local languages (Telugu, Hindi, Marathi) and provided informed consent. The study was conducted in accordance with the tenets of the Declaration of Helsinki. The study was approved by the Ethics Committee for Human Research, LV Prasad Eye Institute, Hyderabad, India.

Parents were administered the package containing the 20-item CarCGQoL questionnaire (described later), and sociodemographic details, including parent's and child's age, gender, parent literacy, employment status, family system (nuclear or joint), number of children with disabilities, duration of the ocular condi-

tion, and approximate monthly family income (in Indian Rupees [INR]). Clinical details such as the type and laterality of the ocular condition were extracted from the child's medical record.

Caregiver's Congenital Glaucoma Quality of Life Questionnaire

The CarCGQoL questionnaire consists of 20 items and is a measure of the caregiver's QoL.[1] The questionnaire provides a measure of overall QoL, and there are no subscales. Broadly, the items target the caregiver's social aspects, emotional well-being, economic, and physical functioning. Parents rated each item on a three-category rating scale consisting of "very much," "moderate amount," and "not at all." The questionnaire was either administered by parents themselves or was administered by a trained interviewer in a face-to-face session in a separate quite room away from the clinic. Given that the questionnaire was developed by us, local language versions were available and translation was not required. Higher CarCGQoL scores indicated better QoL. Previous study of use of the CarCGQoL questionnaire in caregivers of children with PCG demonstrated it to have robust psychometric properties using Rasch analysis.[1]

Statistical Analysis

Descriptive statistics (n, mean, frequency) were used to provide an overview of the characteristics of the study participants that contributed the CarCGQoL scores for Rasch analysis (n = 891). This included all sociodemographic variables fielded in our study. Descriptive statistics were analyzed using the Statistical Package for Social Sciences for Windows, version 19.0 (IBM SPSS, Armonk, NY, USA). Statistical significance was set at $P < 0.05$.

Rasch Analysis

Rasch analysis was applied to test whether the response patterns observed in the data matches the expected theoretical pattern. We first examined the fit of the CarCGQoL questionnaire within each of the three diagnostic groups separately to ensure that each subgroup did not demonstrate any bias in relation to the reliability and person response validity before proceeding to the heterogeneous sample. This step also helped ensure that no validity issues relating to the CarCGQoL questionnaire within each subgroup were overlooked because they otherwise could be hidden in a larger sample. Next the samples were pooled (n = 891) for the analysis. The unit of measurement in Rasch analysis is logit (natural log odds) that is a transformation of a probabilistic value into a linear continuum. We used Winsteps software (version 3.74.0; Chicago, IL, USA) for Rasch analysis by applying the Andrich rating scale model using joint maximum likelihood estimation.[17]

Rasch analysis was performed in a series of steps that included the following: (1) response scale analysis to determine whether the response categories were used by the participants in the order intended, and this was performed by graphically examining whether the thresholds (cross-over point between response categories indicating the point where the likelihood of choosing either response category is the same) advanced monotonically in the category probability curves, that is, as caregivers moved from low to high QoL, were they are more likely to choose "better" response options?; (2) measurement precision (represented by person separation index [PSI]; minimum acceptable value of 2.0, and corresponding reliability; minimum acceptable value of 0.80); (3) unidimensionality (i.e., if all the items measure a single underlying construct of QoL measured by infit mean square statistic with acceptable range of 0.7 to 1.3 and also by principal components analysis of residuals [PCA] whereby eigenvalue >3.0 was considered evidence of a second dimension; for consistency purposes with our CarCGQoL questionnaire development study[1] and also used by other researchers,[18–20] we used a lenient cutoff of 3.0 [instead of eigenvalue < 2.0]); (4) if items match the caregiver's QoL (represented by targeting; ideal <0.5 logits); and (5) differential item functioning (DIF). Detection of DIF in health-related QoL instruments is important for a number of reasons. If DIF is present within a scale, it may be a less-sensitive measure and the content validity of the scale may be affected. The purpose of DIF analysis is to identify items that do not function in the same way across different groups of respondents. In other words, to explore whether bias in responses to the questionnaire exists for certain subgroups. Items are conditioned on some criterion, typically a total score based on a collection of items that measure the same construct. The goal is to determine whether differences in group performance on the item are consistent with group performance differences on the collection of items. DIF examines how group performance on the item varies across levels of the total score; lack of DIF means this relationship is the same across subpopulations. We selected the DIF variables a priori in the present study. DIF was investigated for parent age (split at median), gender, socioeconomic status (high vs. middle/low), laterality of child's ocular condition (unilateral vs. bilateral), type of child's

ocular condition (congenital cataract vs. ROP/corneal conditions, congenital cataract/ROP vs. corneal conditions, ROP vs. congenital cataract/corneal conditions) and language of questionnaire administration (Telugu vs. Hindi/Marathi). For the CarCGQoL questionnaire to be considered stable across groups, we considered DIF to be less than 1.0 logits[21,22] between pairs of diagnostic groups. Using this criterion, we performed DIF assessment for each diagnostic group, as well as for the combined sample.

It has been recommended to use the group for which the scale was normed,[23] the PCG group in the case of the CarCGQoL questionnaire, because the "reference" standard in DIF analysis such that other childhood diseases can be assessed against this group. Given this, we added the data from earlier study of caregiver's QoL of children with PCG to the total group (combined sample) in the present study to detect any DIF-causing items between the PCG sample and the total group.

Results

Participants

Eight hundred ninety-one parents of children with congenital cataract (n = 407 [46%]), ROP (n = 272 [30%]), and blinding corneal disorders (n = 212 [24%]) completed the CarCGQoL questionnaire. Table 1 summarizes the sociodemographic characteristics of the participants. The mean (SD) age of the parents was 28.3 (5.9) years, and most were mothers (n = 680 [76.3%]). Parents of ROP children were significantly younger compared to those of congenital cataract and corneal disorders ($P < 0.0001$). Except for this, other demographic characteristics were comparable across the three groups.

Rasch Analysis Within Each Group

Psychometric properties of the CarCGQoL questionnaire in the three groups are shown in Table 2. The questionnaire demonstrated a well-functioning rating scale across all the three diagnostic groups. There were ordered thresholds between the response categories, indicating that each category has distinct meaning. The categories were evenly spaced (>1.4), illustrating that each category had equal probability to be endorsed by the participants. Some items showed misfit in all three groups. In the congenital cataract group, four items showed misfit (items 2, 3, 19 and 20), and, after an iterative process of item deletion, the remaining 16 items fit the Rasch model. In the ROP group, three items (items 7, 19, and 20) showed misfit and were deleted iteratively. After this step, the remaining 17 items fit the Rasch model. In the corneal disorders group, four items demonstrated misfit (items 3, 15, 19, and 20), and an iterative item removal resulted in 16 items that fit the Rasch model well. Principal component analysis of the residuals showed that the variance explained by the measures was >50% for each group (59.9% for congenital cataract, 58.8% for ROP, as well as corneal disorders), and the unexplained variance explained by the first contrast was <3.0 eigenvalue units, which suggests that the questionnaire was unidimensional for each group. Person separation reliability was adequate across the three groups indicating that the CarCGQoL questionnaire can distinguish among at least three levels of caregivers' QoL in each of the sub-groups. Targeting was 0.39, 0.61, and 0.74 logits in the ROP, corneal disorders, and congenital cataract groups, respectively; ideal targeting was demonstrated by the ROP group.

Rasch Analysis for the Total Group: Differential Item Functioning

Similar to the performance of the rating scale in each diagnostic group, the questionnaire demonstrated a well-functioning rating scale in the total group (combined sample). The thresholds displayed an ordered behavior. Initial analysis of the three groups combined showed that two items (items 19 and 20) misfit the Rasch model considerably. After iterative item removal of these misfitting items, the remaining 18 items fit the Rasch model. The final item difficulties for each group are shown in Figure 1. Unidimensionality of the questionnaire was supported by the PCA of the residuals that showed that variance explained by the measures was > 50% (59.9%), and the unexplained variance explained by the first contrast was <3.0 eigenvalue units, which suggests that the questionnaire was unidimensional for the total group. Person separation reliability was adequate in the total group. Targeting was 0.62 logits indicating that the items were reasonably well suited to the participants (Fig. 2). The most difficult items to endorse by the participants included "experiencing depression," "experiencing anxiety," and "having sleepless nights." By comparison, "feeling guilty" and "experiencing irritability" were the easiest to endorse. Differential item functioning was investigated for the remaining 18 items. However, none of the items demonstrated large DIF (>1.0 logit) by group allocation or any of the variables investigated.

In the DIF analysis of PCG versus the total group, two items ("powerless in facing child's disease" and "lack of interest to pursue any leisure activity")

Table 1. Sample Characteristics

	Combined Sample (n = 891)	Congenital Cataract (n = 407)	ROP (n = 272)	Corneal Disorders (n = 212)
Caregiver demographics				
Age, mean y ± SD	28.3 ± 6.3	29.3 ± 6.6	26.6 ± 5.4	28.7 ± 6.2
Relationship to child, n (%)				
Mother	680 (76)	293 (72)	243 (89)	144 (68)
Father	211 (24)	114 (28)	29 (11)	68 (32)
Marital status, n (%)				
Married	886 (99)	402 (99)	272 (100)	212 (100)
Widowed	5 (1)	5 (1)	0	0
Highest education level achieved, n (%)				
None	114 (13)	84 (21)	10 (4)	20 (9)
Primary	69 (8)	30 (7)	12 (4)	27 (12)
Secondary school	292 (32)	141 (35)	77 (28)	74 (35)
High school	145 (16)	63 (16)	46 (17)	36 (17)
Undergraduate and above	270 (30)	88 (22)	127 (47)	55 (26)
Occupation, father, n (%)				
Daily wage laborer/farmer	339 (38)	178 (44)	71 (26)	90 (42)
Self-employed	228 (26)	112 (28)	71 (26)	45 (21)
Employed for wages	308 (35)	108 (27)	127 (47)	73 (34)
Unemployed	15 (2)	9 (2)	2 (1)	4 (2)
Occupation, mother, n (%)				
Daily wage laborer/farmer	144 (16)	90 (22)	25 (9)	29 (14)
Self-employed	17 (2)	11 (3)	2 (1)	4 (2)
Employed for wages	64 (7)	35 (9)	19 (7)	10 (5)
Home maker	666 (74)	271 (67)	226 (83)	169 (80)
Income level, INR, n (%)				
<5000	361 (41)	190 (47)	80 (29)	91 (43)
5000–10,000	278 (31)	123 (30)	90 (30)	65 (31)
>10,000	250 (28)	92 (23)	102 (38)	56 (26)
Care recipient demographics				
Age, mean months ± SD	36.2 ± 44.8	54.5 ± 50.5	3.7 ± 2.5	42.7 ± 38.9
Median, months	9	48	3	30
Gender				
Male	471 (53)	232 (57)	137 (50)	102 (48)
Female	420 (47)	175 (43)	135 (50)	110 (52)
Affliction, n (%)				
Unilateral	183 (21)	75 (18)	6 (2)	102 (48)
Bilateral	708 (79)	332 (82)	266 (98)	110 (52)
Age at diagnosis, mean months ± SD	31.3 ± 43.9	53.7 ± 50.5	3.9 ± 3.5	23.6 ± 34.9
Duration since diagnosis, mean months ± SD	5.2 ± 14.9	6.9 ± 1.5	0.3 ± 0.6	18.4 ± 24.8
Median, months	0.3	0.3	0.1	7.3
Ordinal position in family				
1	606 (68)	192 (47)	167 (61)	156 (73)
>1	285 (32)	215 (53)	105 (39)	56 (27)
Number of siblings with eye disorder				
1	642 (72)	377 (93)	265 (97)	205 (97)
>1	37 (36)	30 (7)	7 (3)	7 (3)

Values have been rounded off, so percentages may not add to 100% or would slightly exceed 100%.

Table 2. Results of Rasch Analysis of the CarCGQoL Questionnaire in Different Diagnostic Groups

	Congenital Cataract		ROP		Corneal Disorders		Combined Sample	
Parameter	Original Version	Revised Version	Original Version	Revised Version	Original Version	Revised Version	Original Version	Revised Version
No. of items	20	16	20	17	20	16	20	18
Item misfit, n (item nos.)	4 (2,3,19,20)*	0	3 (7,19,20)*	0	4 (3,15,19,20)*	0	2 (19,20)*	0
Reliability	0.85	0.85	0.87	0.85	0.86	0.86	0.86	0.86
Mean person location	0.88	0.74	0.44	0.39	0.67	0.61	0.69	0.62
Principal components analysis (eigenvalue for first and second contrast)	2.2 2.0	2.1 1.9	2.2 1.9	2.0 1.9	2.2 2.0	2.0 1.9	2.1 2.0	2.1 2.0
Differential item functioning, n (notable, >1 logit)	0	0	0	0	0	0	0	0

*Item 2—Because of your child's eye problem, how much anger do you experience?; Item 3—Because of your child's eye problem, how much guilt do you feel?; Item 7—Because of your child's eye problem, how much anxiety do you experience?; Item 15—Because of your child's eye problem, how powerless do you feel in facing your child's eye disease?; Item 19—Because of your child's eye problem, how worried are you about his/her marriage prospects ?; Item 20—How much confidence do you have that your child will be able to see after surgery?

were flagged for large DIF by diagnostic category. Caregivers of children with congenital cataract, ROP, and corneal disorders were more likely to endorse the two items—"powerless in facing child's disease" (1.29 logits) and "lack of interest to pursue any leisure activity" (1.26 logits)—than caregivers of children with PCG.

Discussion

Results from the validation of the CarCGQoL questionnaire in a sample of caregivers of children with congenital cataract, ROP, and corneal disorders revealed the questionnaire to possess acceptable measurement precision (reliability), but with misfitting items for each condition, individually, and as a pooled sample. After iterative item removal, three different versions of the questionnaire (one for each diagnostic condition) emerged from this validation process. Six items demonstrated misfit; four items ("anger," "guilt," "anxiety," "powerlessness in facing child's disease") were dissimilar across the three diagnostic groups, and two items ("marriage prospects" and "see after surgery") demonstrated misfit consistently across the three groups and in the total group (combined sample) of participants. The four items that demonstrated misfit across the three groups were those that addressed the emotional well-being of participants. Given the overlapping item content in the questionnaire that addresses the emotional well-being (albeit not the same aspects such as "depression" and "sleepless nights"), and the focus group discussions we had with caregivers regarding these aspects during the development of this questionnaire previously, we believe that the remainder of the items sufficiently tap into the emotional well-being and thereby the QoL issues of the caregivers of children with ocular conditions. Thus we believe that face validity of the questionnaire is acceptable, thereby providing confidence to future users of the questionnaire.

As noted earlier, two items (Q 19 "marriage prospects" and Q 20 "ability to see after surgery") demonstrated misfit consistently across all the three diagnostic groups. This finding was unexpected given the origin of items from a cohort of caregivers of children with eye disorders, albeit PCG, during the development of the questionnaire. Items misfit for a variety of reasons such as poor construction and, therefore, risk being poorly understood or may be ambiguously worded. Perhaps this was the reason for poor fit of one of these two items (Q20). Whereas

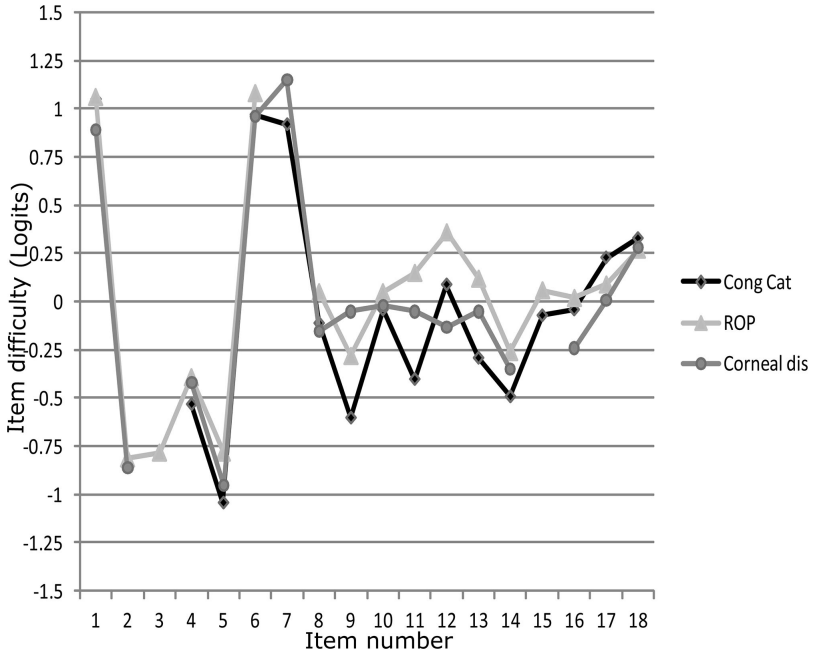

Figure 1. Difficulties (expressed in logits) of the 20-item CarCGQoL questionnaire for caregivers of children congenital cataract, ROP, and corneal disorders after a combined Rasch analysis. Note that items 19 and 20 (misfit) have been removed. Complete description of items can be found in the article by Gothwal et al.[1]

"ability to see after surgery" lacks qualifiers such as see caregiver's face, toys, inanimate objects, faces of familiar people, or any age-appropriate object, these may be considered by users in future studies. Despite these changes, if it continues to demonstrate misfit, then rather than deleting it completely from the questionnaire, it could be analyzed separately such as in a pre-post design study. In case of the other item (Q19), a more likely explanation for the misfit may be related to missing responses to this item (32% of participants) indicating that most of the caregivers never thought of these issues. Given the high proportion of missing responses to this item, we considered it appropriate to delete it. Health-related assessment tools that lack measurement equivalence across population subgroups can result in flawed research and erroneous clinical decisions.

We investigated whether the pooled sample respond to the items of the CarCGQoL questionnaire in the same way as the reference group (caregivers of children with PCG). This is an important prerequisite to determine whether using the CarCGQoL questionnaire is justified in caregivers of congenital cataract, ROP, and corneal disorders and whether their CarCGQoL scores can be interpreted in the same way as in caregivers of children with PCG. The pooled sample responded significantly differently to two items ("powerless in facing child's disease" and "lack of interest to pursue any leisure activity") out of a total of 20 items (10%), when compared with the reference group. Regardless of the level of their QoL, the pooled sample was more likely to endorse these two items compared to the caregivers of PCG. This would mean that if a caregiver of a child with congenital cataract, ROP, and corneal disorders has high QoL, yet endorses these two items, the questionnaire score could be lower than for a caregiver of a child with PCG with lower QoL. This perhaps suggests that not all of the items in the CarCGQoL questionnaire assess caregiver's QoL equally across those with PCG versus different childhood ocular conditions. Therefore caution should be exercised when comparing the results of the QoL of caregivers of children with PCG and other childhood ocular conditions, or when pooling the data in a single analysis of CarCGQoL questionnaire. Although problems with translation of items in a questionnaire may be one of reasons for an item to be flagged for DIF, we believe that this may not be the case in our study given that none of these items demonstrated DIF by language. Even though the CarCGQoL questionnaire was tested to ensure that the questions are worded properly during the development process, there still remains the possibility that certain groups of caregivers not previously examined may have different interpretations. However, in the present study, all the caregivers were provided with a choice of language among the

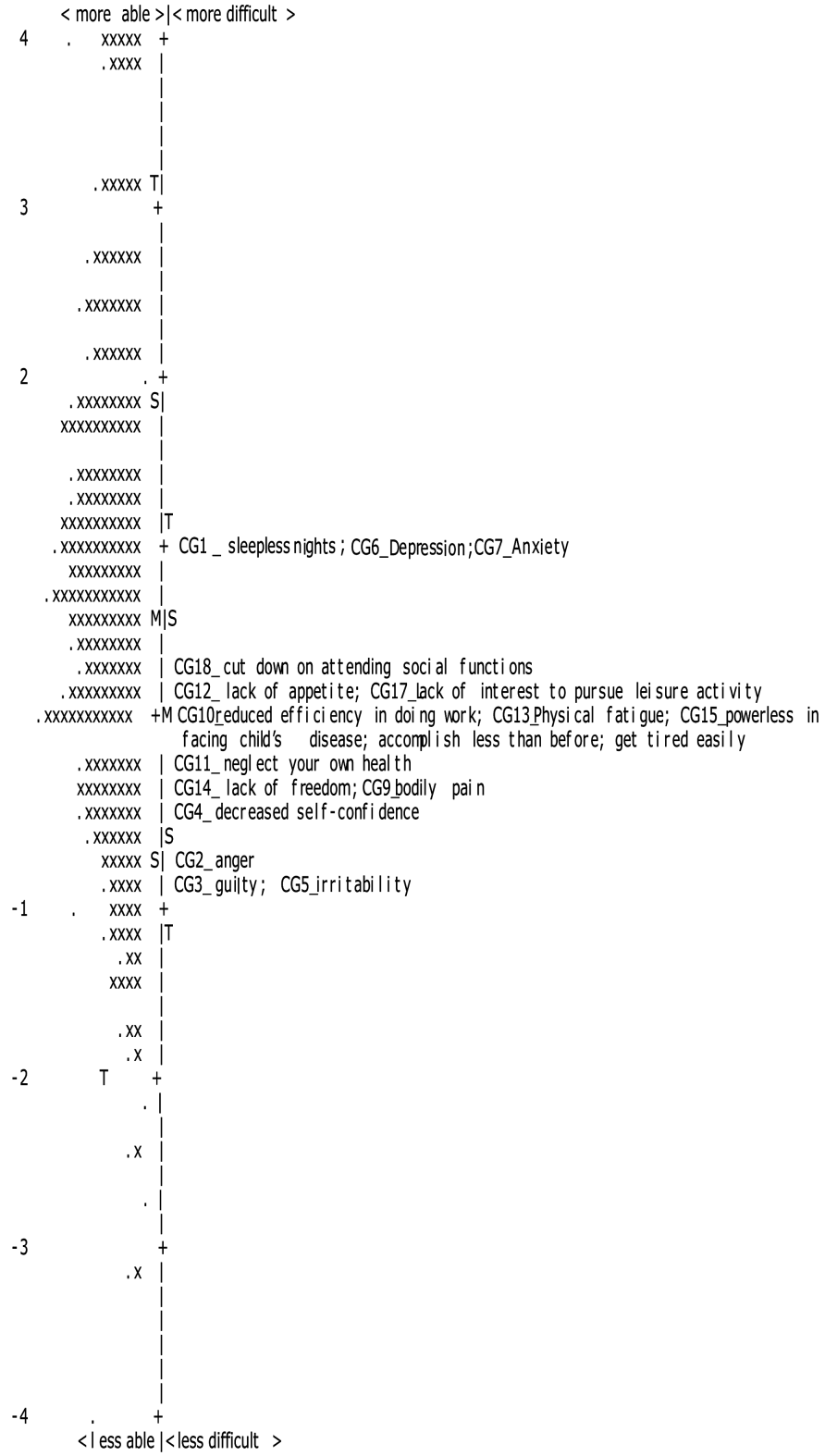

Figure 2. Person-item map for the CarCGQoL questionnaire (n = 891) in a combined sample of caregivers of children with congenital cataract, ROP, and corneal disorders. The *vertical line* represents the measure of the quality of life, in logit units. Participants appear in ascending order of ability (on the left hand side of the map) while the items appear in ascending order of difficulty (on the right hand side of the map). Alongside each item is also indicated its number as in the 20-item original CarCGQoL questionnaire. Item names have been abbreviated to fit the space and the correct description of items can be found in the article by Gothwal et al.[2] Each *x* represents four participants, and each *period* represents one to three participants. M, mean; S, 1 SD from the mean; T, 2 SD from the mean. By convention, the mean item difficulty is set at 0 logits (indicated with *M*). Accordingly, mean quality of life of participants is indicated with M.

three available and had the opportunity to ask for clarifications if they were confused. Although we do not have a ready explanation for the occurrence of DIF in the present study, the likelihood of poor wording being the primary cause of the DIF found between the PCG group and other childhood ocular conditions appears remote, and rewording or removal of items is probably unwarranted. Although the deletion of these two DIF-causing items did not appear to have an appreciable impact on the measurement precision (0.84 logits) and targeting (0.62 logits), these two items are still relevant because they tap into social functioning and may be providing little QoL-related information for one cohort but valuable information for another. Replication is clearly needed to support this and all of the findings presented here. Should it be necessary to compare the caregiver's QoL between PCG and other childhood ocular conditions, special attention should be paid to these two items that exhibit DIF.[24,25]

The CarCGQoL questionnaire demonstrated acceptable measurement precision (reliability > 0.80), which indicates that it can separate at least three distinct groups of caregivers with perceived QoL, thus supporting its use as a measure.[26] Furthermore, our results showed that the CarCGQoL items form a unidimensional scale within each caregiver group, which confirms that the items measure one underlying construct and that the summation of individual item scores to create a total score is justified in caregivers of children with corneal disorders, ROP, and congenital cataract. Taken together, these results support the validity of the CarCGQoL questionnaire for each diagnostic group tested, thus supporting its generic measurement properties.

Our results showed that, in accordance with the item hierarchy in the original scale among caregivers of children with PCG, items such as "experiencing depression," "experiencing anxiety," and "having sleepless nights" were most the difficult and "feeling guilty" and "experiencing irritability" were the easiest to endorse by the participants.[1] The intermediate items showed some, albeit minor, variation in item hierarchy when compared with the PCG sample. As can be seen from Table 2, targeting of item difficulty to person ability was ideal only for the ROP group (0.39 logits) but was still within the acceptable range (<1.0 logit) for the other two groups (0.61 and 0.74 logits) and for the combined sample (0.62 logits). However, Figure 2 shows that there were many redundant items, as well as a lack of "hard" items to target the "most able" participants (at the top). This suggests that the questionnaire has relatively better precision to reliably detect differences and changes in caregivers with worse QoL as compared to those with better QoL. The results of the present study regarding the optimal psychometric properties of CarCGQoL questionnaire across different diagnostic groups are perhaps not surprising given that the psychological, financial, and social issues that a caregiver has to face when his/her child is initially diagnosed with an eye disorder are similar, be it PCG, congenital cataract, ROP, or corneal disorders. Nonetheless, as mentioned earlier, psychometric validity in another patient group cannot be assumed and needs to be tested and demonstrated as has been done in the present study.

There are a few limitations in our study. Caregivers of children with ROP were significantly younger compared to those of congenital cataract and corneal disorders and reported comparatively much worse QoL. Given this, the possibility that at least a portion of the variations in the caregiver's QoL associated with age difference may be the result of DIF rather than actual difference in QoL cannot be excluded. The samples analyzed in our study are based on a combination of strategic and convenience sampling procedures. It is therefore difficult to draw general conclusions from the findings of the study to a larger representative sample. However, the variations in diagnostics and the wide range of demographics in combination with rather large sample sizes may contribute information that may be clinically relevant for pediatric ophthalmologists.

It should be acknowledged that, the responsiveness of the questionnaire to intervention has not been assessed as yet. Nevertheless, the results of the present study highlight the importance of better understanding the role of child's ocular condition on caregiver's QoL and suggest areas of future research. For example, the impact of chronic childhood ocular conditions that are not life-threatening versus those that are life-threatening (such as retinoblastoma), on the caregiver's QoL can be attempted in future studies.

Although our sample refers to a very specific population of caregivers of children with congenital cataract, ROP, and corneal disorders, we believe that several challenges that are encountered by this group are also shared with other difficult caregiving scenarios. As such, further research is warranted to replicate these findings among other caregiver populations, for example, in caregivers of children with syndromic ocular conditions, retinoblastoma, and more.

In conclusion, this analysis of the cross-diagnostic validity of the CarCGQoL questionnaire shows that care must be taken when data from different pediatric ocular conditions are pooled given the presence of DIF between the reference group (PCG) and the pooled sample. This suggests that when evaluating

the impact of interventions on the caregiver's QoL in a pooled sample, cross-diagnostic DIF must be taken into account. However, it must be emphasized that before any recommendations can be made for modifying the CarCGQoL questionnaire based on DIF, the results found here must be replicated with other pediatric ocular conditions as well. This study suggests that caregivers of children with different pediatric conditions do not define QoL in exactly the same manner, and research involving QoL that uses different pediatric conditions cannot ignore this important issue. The continuing misfit of some items in three different diagnostic conditions resulted in three different versions of the CarCGQoL, but these condition-specific versions are valid and reliable for measuring the QoL of caregivers of children with chronic pediatric ocular conditions. It is an easy instrument to administer and can be completed by the caregiver before the first meeting with the ophthalmologist, which is an advantage when having to deliver intervention in time constrained health care service. This is the first attempt to test the cross-diagnostic validity of the CarCGQoL questionnaire, and we hope that there will be increased research in this area.

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References

1. Gothwal VK, Bharani S, Mandal AK. Quality of life of caregivers of children with congenital glaucoma: development and validation of a novel questionnaire (CarCGQoL). *Invest Ophthalmol Vis Sci*. 2015;56:770–777.
2. Gothwal VK, Bharani S, Mandal AK. Impact of Surgery on the Quality of Life of Caregivers of Children with Congenital Glaucoma. *Ophthalmology*. 2016;123:1161–1162.
3. Kuhlthau K, Kahn R, Hill KS. The well-being of parental caregivers of children with activity limitations. *Matern Child Health*. 2010;14:155–163.
4. Adelman RD, Tmanova LL, Delgado D, Dion S, Lachs MS. Caregiver burden: a clinical review. *JAMA*. 2014;311:1052–1060.
5. Lovell B, Wetherell MA. The cost of caregiving: endocrine and immune implications in elderly and non elderly caregivers. *Neurosci Biobehav Rev*. 2011;35:1342–1352.
6. Klassen AF, Gulati S, Granek L, et al. Understanding the health impact of caregiving: a qualitative study of immigrant parents and single parents of children with cancer. *Qual Life Res*. 2012;21:1595–1605.
7. Medalie J. The caregiver as the hidden patient. In Kahana E, Biegel DE, Wykle M (eds.), *Family caregiving across the lifespan*. Thousand Oaks, CA: Sage Publication, Inc.; 1994:312–330.
8. Stein RE, Riessman CK. The development of an impact-on-family scale: preliminary findings. *Med Care*. 1980;18:465–472.
9. Reine G, Lancon C, Simeoni MC, Duplan S, Auquier P. Caregiver burden on relatives of persons with schizophrenia: an overview of measure instruments. *Encephale*. 2003;29:137–147.
10. Pinquart M, Sorensen S. Differences between caregivers and noncaregivers in psychological health and physical health: a meta-analysis. *Psychology and Aging*. 2003;18:250–267.
11. Higginson IJ, Gao W, Jackson D, Murray J, Harding R. Short-form Zarit Caregiver Burden Interviews were valid in advanced conditions. *J Clin Epidemiol*. 2010;63:535–542.
12. Pinquart M, Sorensen S. Differences between caregivers and noncaregivers in psychological health and physical health: a meta-analysis. *Psychol Aging*. 2003;18:250–267.
13. Linder M, Chang TS, Scott IU, et al. Validity of the visual function index (VF-14) in patients with retinal disease. *Arch Ophthalmol*. 1999;117:1611–1616.
14. Parrish RK, 2nd, Gedde SJ, Scott IU, et al. Visual function and quality of life among patients with glaucoma. *Arch Ophthalmol*. 1997;115:1447–1455.
15. Pesudovs K, Caudle LE, Rees G, Lamoureux EL. Validity of a visual impairment questionnaire in measuring cataract surgery outcomes. *J Cataract Refract Surg*. 2008;34:925–933.
16. Dallmeijer AJ, de Groot V, Roorda LD, et al. Cross-diagnostic validity of the SF-36 physical functioning scale in patients with stroke, multiple sclerosis and amyotrophic lateral sclerosis: a study using Rasch analysis. *J Rehabil Med*. 2007;39:163–169.

17. Andrich DA. A rating scale formulation for ordered response categories. *Psychometrika*. 1978;43:561–573.
18. Hwang JE. Promoting healthy lifestyles with aging: development and validation of the Health Enhancement Lifestyle Profile (HELP) using the Rasch measurement model. *Am J Occup Ther*. 2010;64:786–795.
19. Marella M, Gothwal VK, Pesudovs K, Lamoureux E. Validation of the visual disability questionnaire (VDQ) in India. *Optom Vis Sci*. 2009;86:E826–835.
20. Smith EV, Jr. Detecting and evaluating the impact of multidimensionality using item fit statistics and principal component analysis of residuals. *J Appl Meas*. 2002;3:205–231.
21. Wright BD, Douglas GA. *Best test design and self-tailored testing*. Chicago, IL: Statistical laboratory, Department of Education, University of Chicago; 1975.
22. Wright BD, Douglas GA. *Rasch item analysis by hand*. Chicago, IL: Statistical Laboratory, Department of Education, University of Chicago; 1976.
23. Dorans NJ, Schmitt MP. *Constructed response and differential item functioning: A pragmatic approach*. Princeton: ETS; 1991.
24. Crane PK, Cetin K, Cook KF, Johnson K, Deyo R, Amtmann D. Differential item functioning impact in a modified version of the Roland-Morris Disability Questionnaire. *Qual Life Res*. 2007;16:981–990.
25. McHorney CA, Fleishman JA. Assessing and understanding measurement equivalence in health outcome measures. Issues for further quantitative and qualitative inquiry. *Med Care*. 2006;44:S205–210.
26. Smith EV, Jr. Evidence for the reliability of measures and validity of measure interpretation: a Rasch measurement perspective. *J Appl Meas*. 2001;2:281–311.