

Quality of Life of Caregivers of Children With Congenital Glaucoma: Development and Validation of a Novel Questionnaire (CarCGQoL)

Vijaya K. Gothwal,¹ Seelam Bharani,¹ and Anil K. Mandal²

¹Meera and L B Deshpande Centre for Sight Enhancement, L V Prasad Eye Institute, Hyderabad, India

²Jasti V Ramanamma Children's Eye Care Centre, L V Prasad Eye Institute, Hyderabad, India

Correspondence: Anil K. Mandal, Jasti V Ramanamma Children's Eye Care Centre, L V Prasad Eye Institute, Kallam Anji Reddy Campus, L V Prasad Marg, Banjara Hills, Hyderabad 500034, Andhra Pradesh, India; mandal@lvpei.org.

Submitted: October 20, 2014
Accepted: December 26, 2014

Citation: 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. DOI:10.1167/iops.14-15905

PURPOSE. We developed and validated an instrument to measure the quality of life (QoL) of caregivers of children with primary congenital glaucoma (PCG): the Caregiver's Congenital Glaucoma Quality of Life (CarCGQoL) questionnaire.

METHODS. A total of 70 caregivers of children with PCG under follow-up at a tertiary eye care center in India participated in a series of focus groups, and pre- and pilot testing phases in the development of the 45-item CarCGQoL questionnaire. Subsequently, this instrument was administered to 111 caregivers of children with PCG awaiting glaucoma surgery. Response to each item was rated on a four-category scale. Rasch analysis was used to validate the instrument's psychometric properties, such as unidimensionality (by principal components analysis [PCA] of residuals), item fit to model, measurement precision (by person separation [PS]), response category performance, differential item functioning (DIF), and targeting of items to participant's QoL.

RESULTS. Categories required reorganization resulting in a shortened rating scale of 3 categories. The instrument showed misfit to the Rasch model and lacked unidimensionality. Deleting 15 items (obtained through PCA of residuals) restored unidimensionality, but additional items misfit, necessitating item reduction. In addition, 2 items showed DIF and were deleted. Finally, a 20-item CarCGQoL instrument showed good fit and unidimensionality. Targeting was good (0.69 logits) and PS was 2.51, indicating good measurement precision.

CONCLUSIONS. We have developed a psychometrically robust 20-item caregiver derived questionnaire, the CarCGQoL, that is a valid and reliable measure of QoL of caregivers of children with PCG. Given its brevity, it is quick and easy to administer in the clinic, and has potential for use as an outcome measure in clinical trials of treatment of PCG.

Keywords: caregiver, quality of life, congenital glaucoma, questionnaire

Primary congenital glaucoma (PCG) results from developmental abnormality of the trabecular meshwork as well as of the anterior chamber angle of the eye. It is found commonly between birth to 3 years, with the majority being diagnosed during the first year of life.¹ Congenital glaucoma is responsible for 4% of childhood blindness² and the prevalence of PCG has been estimated to be 1 per 3300 births in the Indian state of Andhra Pradesh^{1,3} as opposed to 1 per 10,000 births in the Caucasian populations.⁴ It more often is familial in nature in Indian than it is in Caucasian populations.⁵⁻⁷ Evaluation of severity of PCG relies principally on the level of the IOP, extent of corneal enlargement, and corneal transparency.⁸ It is primarily a surgical disease, and the goal of surgery is to reduce IOP and restore corneal transparency.^{9,10} Several retrospective studies conducted worldwide have evaluated the benefits of surgical intervention for patients with PCG.⁹⁻¹⁸ Although untreated PCG often leads to blindness, a proportion of treated children with severe forms of the disease also have poor visual outcomes.^{10,13}

Studies of management of PCG typically focus on clinical measures of efficacy related to control of IOP and restoration of corneal transparency.⁹⁻¹⁸ Children with PCG require lifelong

follow-up for periodic assessment and monitoring of IOP,¹⁰ and this places huge demands on the part of the family members. Especially in the first few months and years after diagnosis and surgery, parents are faced with the fact that PCG presents a serious threat to their child's visual development. Given that PCG is a chronic disorder that manifests commonly in the newborns and infants, the responsibility of providing constant care to children with the disorder falls on the caregivers (generally their parents). This takes a tremendous toll on the physical and emotional health of the caregivers. More importantly, a child's illness is likely to affect a parent's quality of life (QoL).¹⁹ Quality of life as defined by the World Health Organization (WHO) is "an individual's perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards, and concerns."²⁰ Given this, it is important to know how satisfied or bothered people are by important aspects of life. Therefore, in the context of PCG, new approaches that include paying attention to the caregiver's QoL (measure of physical, psychological, and social well-being) may be helpful in complementing the traditional measures of visual and surgical outcomes because an impaired QoL not only adversely

affects the caregiver's personal health, but also may impact the quality of care that they provide to the child with PCG. As noted earlier, while several studies have investigated the clinical outcomes in patients with PCG, the impact of the diagnosis, management, and the care process thereof on the caregiver's QoL has remained largely unexplored. Common approaches to assessing the impact of caring for a chronically ill child have included measuring the impact of the child's illness on the caregiver's well-being and QoL (for example, Pediatric Asthma Caregiver's QoL questionnaire,²¹ the Caregiver Oncology QoL questionnaire²²), evaluating the effect of the child's illness on family resources, asking caregivers open-ended questions about family strain, burden of care that the condition possesses, amount of physical stress associated with caring for the child, and the amount of time that is required to care for the child's medical, physical, and social needs.²³ This latter approach was followed in a study that assessed caregiver burden in PCG.²⁴ Using the caregiver burden questionnaire (CBQ) and the patient-health questionnaire, the investigators reported that the caregivers of children with PCG have significant emotional and psychological burden, and that moderate to severe depression may be present in one-third of the caregivers.²⁴ To our knowledge, a QoL questionnaire for caregivers of children with PCG has not yet been developed. The aim of this study was to develop an instrument – the Caregivers Congenital Glaucoma Quality of Life Questionnaire (CarCGQoL), capable of estimating the caregiver's QoL that could provide insights into the challenges of the caregiver's everyday lives while caring for their child with PCG. The measure can be completed while parents/caregivers are waiting to be seen by the ophthalmologist, and has been designed to complement diagnostic information and other clinical data by assessing the experiences and feelings of parents/caregivers and the degree to which their child's PCG affects their QoL.

PATIENTS AND METHODS

The study was approved by the Ethics Committee for Human Research at the L V Prasad Eye Institute (LVPEI), Hyderabad, India, and followed the tenets of the Declaration of Helsinki. Each parent/caregiver gave informed consent.

The development and validation of the questionnaire followed four distinct phases. Item generation (phase I) based on literature review, focus group discussions (FGDs), and consultation with the experts (pediatric glaucoma specialists), and this was followed by item reduction/refinement (phase II), piloting, or cognitive debriefing (phase III), and, finally, validation of the CarCGQoL questionnaire (phase IV) with caregivers of children with PCG. For all four phases of the study, caregivers of children with PCG were enrolled. All children were diagnosed to have PCG and had either undergone glaucoma surgery (for phases I–III of the questionnaire development) or were scheduled for surgery at our center (for phase IV). Databases of children with PCG who underwent glaucoma surgery by a single surgeon (AKM) at the LVPEI between May 2004 and May 2006, and were under follow-up care were used for recruitment in the first and third phases of the study. In phase I, 30 eligible caregivers were invited to participate in FGDs (6 per group). Focus groups are a popular method to explore the experience of QoL from the perspective of a target population that help in conceptualizing and operationalization of important constructs of QoL, thereby, informing identification of themes. They are especially useful for exploration of phenomena and experiences that are generally poorly understood, when the literature or expert opinion may not capture all relevant issues pertinent to the

questionnaires intended for the target population.^{25,26} Put simply, focus groups are a method where the developers of questionnaires gain an “insider perspective” from those for whom the questionnaire is intended to be used.^{27,28} For phase I of the study, caregivers were invited when they accompanied their child for follow-up care to the LVPEI between December 2006 and May 2008 (i.e., at least 6 months after surgery). However, 5 caregivers participated in one–one qualitative interviews as they could not attend the FGDs (because of logistical reasons). In phases III and IV, 40 and 111 caregivers participated, respectively. The child's primary caregiver was the person who was most responsible for the day-to-day care of the child and spoke Hindi, Telugu, or English. Before each FGD, information about the purpose of the study and confidentiality were provided and informed consent was obtained. No compensation was offered.

Phase I: Item Generation

We performed a focused literature review to assess the content of some of the generic QoL questionnaires (SF-36) and caregiver's QoL questionnaires that have been developed for chronic pediatric medical conditions, for example, asthma²¹ and cancer,²² so as to understand the style of item wording (direct versus indirect) and response categories. However, we did not find any of these questionnaires useful in their entirety for our purpose, so we decided to develop our own questionnaire. The content for the CarCGQoL questionnaire was derived from 5 FGDs consisting of a total of 30 caregivers (mean age, 24.6 ± 9.8 years; 80% mothers) supplemented by qualitative semistructured interviews with 5 caregivers and 3 experts (pediatric glaucoma specialists). The topics for the FGDs were informed by Conceptual Model for QoL proposed by Ferrans²⁹ that identifies four domains: health and functioning, psychological/spiritual, social and economic, and family. All FGDs and qualitative semistructured interviews were led and moderated by an experienced qualitative researcher with a graduate degree (PhD) helped by two research assistants (RAs) so as to identify issues (such as health and functioning, psychological/spiritual, social and economic, and family) that parents/caregivers considered to be affected because of the diagnosis of PCG, need for surgery and glasses for their child, and life-long follow-up. Each FGD lasted 1 to 2 hours during which participants gave their opinions of the issues, disagreed among themselves, agreed, discussed controversial issues, and in short, intensely participated in the discussions. During the FGDs, the RAs made running notes of the issues raised by the participants independently and the exact words/phrases used in local language were noted such that the essence was captured. At the end of each FGD, the moderator collated their notes/findings and summarized them aloud (ensuring anonymity). Any disagreements were resolved immediately, and a list of issues that was agreed upon by the whole group was finalized. The same procedure was followed for all the FGDs. Similarly, semistructured interviews were conducted with some of the caregivers and the researcher made notes of the responses to the several leading questions regarding the impact of their child's PCG (its diagnosis, economic concerns, visual and surgical outcomes, concerns regarding lifelong follow-up) on their life and the concerns that they may have about their child's PCG.

The sentences and phrases were independently entered into an Excel database by the RAs, and were jointly reviewed by them and the moderator. Unique individual meaningful statements or phrases regarding the impact of PCG on the caregiver's QoL were converted into questions/items. However, specific statements that were likely to discriminate between caregivers based on their sex, and socioeconomic or educational status were not included. We borrowed two items from SF-36. The

themes from the semistructured interviews were not, however, distinct from those in the FGDs. Statements with common themes were pooled together and this was done for each FGD, and a final list of 85 items was prepared.

Phase II: Item Reduction/Refinement

Following the generation of the statement pool from each of the FGDs, the list was reanalyzed and redundancies were removed through item reduction. In this phase, the experts (pediatric glaucoma specialist) and researchers independently reviewed all the statements. The first phase of item reduction consisted of a "macro level" removal of items relating to content themes that were not appropriate for inclusion in the CarCGQoL; a self-report measure of caregiver's QoL ($n = 15$). Decisions regarding the removal of all items within a content theme (e.g., impact of PCG on caregiver's QoL) were informed by the literature and by the research team including the clinical opinion of the treating ophthalmologist (pediatric glaucoma specialist) involved in the project. Item quality was assessed by the team, and items with poor reading ease and/or potential ambiguity were deleted from the pool. Ten statements were reworded following expert review and then wording of the remaining items was adapted to maximize consistency of item wording and response format. Items then were formatted as the draft CarCGQoL questionnaire consisting of 45 items. The questionnaire was initially developed in English (although this version was not used in the present study given that none of the caregivers opted for it) and subsequently was translated and back-translated into three local languages, that is, Telugu, Marathi, and Hindi using standard accepted procedures.

Phase III: Cognitive Debriefing

This technique is used commonly to determine whether the meaning of an item, as intended by the questionnaire developer, is consistent with the participant's interpretation of that item.³⁰ We conducted cognitive debriefing to explore caregiver's views on the comprehensibility, acceptability, relevance, and answerability of the draft CarCGQoL questionnaire to inform its refinement. Participants consisted of a purposive sample of 40 caregivers of children with PCG having differing severities (mild to moderate) and laterality of PCG (unilateral and bilateral). Each participant completed the CarCGQoL in the presence of a researcher (VKG), who elicited comments about specific items, type of response category format (frequency versus severity), the desirable number of categories, and ascertained their opinions about the questionnaire in general (e.g., content, layout, length). At this stage of development, participants chose the severity form of response categories ("not at all" to "very much") over frequency. In addition, this stage informed changes to the wording of retained items to increase consistency of item wording and minimize participant burden. For example, in the initial version of the CarCGQoL, the word "experience" appeared in many of the statements (Because of your child's glaucoma how much do you experience . . .). The word "experience" was removed from the statements and a common phrasing question was used for 30 of the 45 items. This change occurred in response to suggestions that the repetition in wording of items was irritating and disturbing to the respondents.

Phase IV: Validation of the CarCGQoL

For this phase of the study, we recruited 111 caregivers of children with PCG who were scheduled to undergo glaucoma surgery. The 45-item CarCGQoL was self-administered by 47 caregivers (42%) and was interviewer administered for 64

TABLE 1. The 20-Item CarCGQoL

Item No.	Item Description
1	Sleepless nights
2	Anger
3	Guilty
4	Decreased self-confidence
5	Irritability
6	Depression
7	Anxiety
8	Get tired easily
9	Bodily pain
10	Reduced efficiency in doing work
11	Neglect your own health
12	Lack of appetite
13	Physical fatigue
14	Lack of freedom
15	Powerless in facing child's disease
16	Accomplish less than before
17	Lack of interest to pursue any leisure activity (like watching TV, spending time in neighbor's house)
18	Cut down on attending social functions
19	Your child's marriage prospects
20	Your child will be able to see after surgery

Framing questions for items 1 to 18: Because of your child's glaucoma how much do you experience the following. . . . Framing question for item 19: How worried are you about. . . (item). Framing question for item 20: How confident are you about. . . (item). Response categories for items 1 to 19: Very much (0), A little to moderate amount (1), Not at all (2), Not applicable (3). Response categories for item 20: Very much (2), A little to moderate amount (1), Not at all (0), Not applicable (3).

(58%). The outcome of the validation of the 45-item CarCGQoL questionnaire (resulting in formation of the final 20-item CarCGQoL questionnaire) that was administered to 111 caregivers is presented in the Results section. Table 1 consists of the item content of the 20-item CarCGQoL questionnaire.

Data Collection: Demographic and Clinical Data

Sociodemographic data, including age and sex of the child, age of the caregiver, relationship to the child with PCG, number of children in the family, ordinal position of the child with PCG, monthly family income, age at diagnosis of PCG, and family history of PCG, were collected from the caregivers alongside the responses to CarCGQoL questionnaire. Clinical data, such as laterality, type of PCG, severity of PCG, were ascertained from the medical records. The severity index proposed by Panicker et al.⁸ was used to grade the severity of the PCG.

Statistical Analysis

Rasch analysis was conducted using the Andrich rating scale model³¹ with Winsteps software version 3.74.0 (Chicago, IL, USA).³² The details of Rasch analysis have been detailed in earlier reports.^{33,34} In brief, Rasch analysis provides an estimate of person ability and item difficulty along a common measurement continuum.³⁵ Ordinal measures are transformed into linear continuous measures of person ability (QoL in present case) and item difficulty, which are expressed in logits. The stepwise analytic approach and criteria for evaluating each psychometric property have been described previously by us.³⁶⁻³⁸ Therefore, we report them in brief here. Five fundamental indicators of Rasch analysis were used to evaluate the CarCGQoL questionnaire. These included: (1) item fit (using infit mean square values of 0.7-1.3 and items with

TABLE 2. Demographic and Clinical Characteristics of the 111 Participants of Validation Phase

Characteristic	Proportion
Caregiver	
Age, mean $y \pm$ SD	25.3 \pm 4.3
Sex, <i>n</i> (%)	
Male	17 (15)
Female	94 (85)
Marital status, <i>n</i> (%)	
Married	110 (99)
Widowed	1 (1)
Education, <i>n</i> (%)	
None	15 (13)
1-5 y	12 (11)
6-10 y	40 (36)
>10 y	44 (40)
Relationship to child, <i>n</i> (%)	
Mother	96 (86.5)
Father	15 (13.5)
Occupation, <i>n</i> (%)	
Farmer	10 (9)
Laborer	12 (11)
Homemaker	71 (64)
Teacher	7 (6)
Tailor	2 (2)
Other	8 (7)
Unemployed	1 (1)
Income level, INR, <i>n</i> (%)	
<5,000	49 (44)
5,000-10,000	42 (38)
>10,000	20 (18)
Care recipient	
Age, mean mo \pm SD	5.7 \pm 8.4
Median age, mo	4
Sex, <i>n</i> (%)	
Male	64 (58)
Female	47 (42)
Affliction, <i>n</i> (%)	
Unilateral	27 (24)
Bilateral	84 (76)
Severity of PCG,* no. of eyes† (%)	
Mild	75 (38.5)
Moderate	77 (39.5)
Severe	43 (22.0)
Type of PCG, <i>n</i> (%)	
Congenital (less than 1 mo)	81 (73)
Infantile (1-3 y)	27 (24)
Juvenile (>3 y)	3 (3)
Age at diagnosis (d), <i>n</i> (%)	
<30	68 (61)
30-90	33 (30)
>90	10 (9)
Ordinal position in family, <i>n</i> (%)	
1	56 (50)
>1	55 (50)
Number of children with glaucoma, <i>n</i> (%)	
1	106 (96)
>1	5 (4)
Presenting symptoms, no. of eyes† (%)	
Blue eyes	54 (28)
Big eyes	16 (8)
Watering	10 (5)
Photophobia	8 (4)
Vision loss	2 (1)
More than one symptom	76 (39)

TABLE 2. Continued

Characteristic	Proportion
Referred by pediatrician	29 (15)

INR, Indian rupees.

* Classification as proposed by Panicker et al.⁸ (see text for details).

† 195 eyes.

values outside this range were considered misfits that were deleted in iterative manner) and unidimensionality using principal components analysis of residuals (PCA); (2) item difficulty; (3) targeting (ideal targeting exists when the mean of items equals the mean of persons—the greater the difference in means the poorer is the targeting); (4) person separation index (PSI)—minimum acceptable value was 2.0 (person reliability 0.80); and (5) differential item functioning (DIF) – whether sample subgroups respond differently to certain items despite possessing equal amounts of the underlying trait. We selected the DIF variables a priori in this study and investigated DIF for caregiver's age (split at median; <25 vs. \geq 25 years), child's age (split at median; <4 vs. \geq 4 months), sex (male versus female), socioeconomic status (SES; high versus low), order of birth (first versus others), number of children with glaucoma (1 vs. \geq 1), mode of questionnaire administration (self- versus interviewer-administered), age at diagnosis (<30 vs. \geq 30 days). When an item was found to exhibit notable DIF (>1.0 logit^{39,40}), deletion was considered, especially if removal improved the performance of the overall model.⁴¹ Given that we used three language versions of the CarCGQoL we examined the cross-cultural/linguistic validation of the instrument through an assessment of DIF. Each participant was administered the instrument in only one language, so we used the nested study design that provides DIF for item effects within language group in DIF testing. Overall differences in language difficulty will be shown by differences in average ability for the participants being tested in each language, and we used the 1-way ANOVA to investigate this difference. Statistical significance was set at $P < 0.05$.

In addition, Rasch analysis enables to assess whether the participants have used the rating scale (using the category probability curves) as was envisaged by the developers and in case of deficiencies then the response categories may need reordering. A participant who has high QoL will have positive logit score and vice versa. While this is valid for 31 items, it is just the opposite for the remaining 15 items. Therefore, we reverse coded the categories for these 14 items, such that a positive logit score will indicate high QoL across all the items. The Statistical Package for Social Sciences (v. 19.0; SPSS, Inc., Chicago, IL, USA) was used to perform descriptive statistics to characterize the participants' sociodemographic, and clinical and CareCGQoL data. Statistical significance was set at $P < 0.05$.

RESULTS

Sociodemographics and Clinical Characteristics of the Participants

We administered the 45-item CarCGQoL questionnaire to 111 caregivers. The mean age of the caregivers was 25.3 years (SD, 4.3 years; range, 18-41). Most of these were mothers ($n = 96$, 86.5%), had at least >5 years of education ($n = 84$, 76%), and were homemakers ($n = 71$, 64%). A little less than half of the caregivers ($n = 49$, 44%) belonged to a lower SES and 61% ($n = 68$) of the children were diagnosed to have PCG within 1 month of their birth. The demographic characteristics of the caregivers are provided in Table 2.

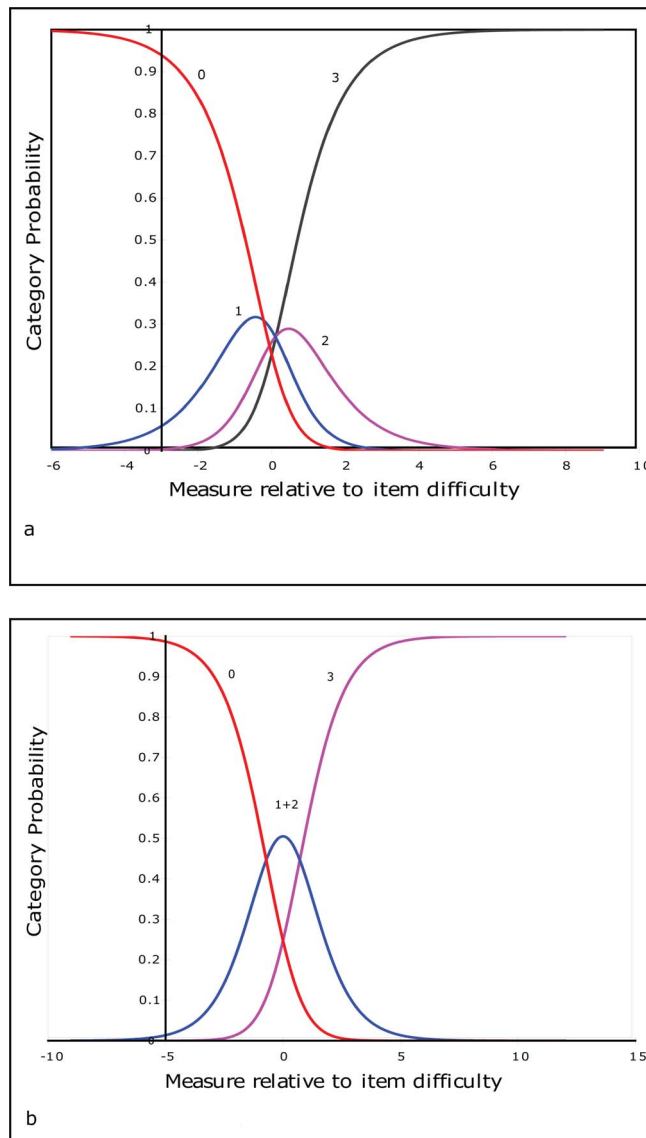


FIGURE. (a) Rasch model category probability curves for all items together in the CarCGQoL questionnaire showing the likelihood that a participant with a particular QoL will select a category. The *scale* (*x*-axis) from -6 to $+10$ symbolizes the latent trait of QoL. The *y*-axis represents the probability of category being selected. Response categories were 0, “very much”; 1, “moderate amount”; 2, “a little”; and 3, “not at all.” For any given point along this scale, the category most likely to be chosen by a participant is shown by the category curve with the highest probability. At no point was category 2 the most likely to be chosen, resulting in disordered thresholds. Thresholds represent boundaries along the scale where the probability of a response category being chosen changes from one to the next. (b) Combining categories 1 and 2, and thereby reducing the number of categories from 4 to 3, repaired the disordered thresholds for the category probability curves seen in (a).

Psychometric Validation of the CarCGQoL Questionnaire

We commenced our analysis with an assessment of the rating scale (response categories) to determine if the participants had used the categories as was conceptualized by us. Figure a illustrates the category probability curves (CPC) of an item in its 4-point original form showing disordered thresholds necessitating collapsing of categories. The CPC plots QoL on

the *x*-axis against the probability of endorsing each response category on the *y*-axis. Threshold refers to the point between two adjacent response categories, for example, 1 and 2, where either response (1 or 2) has equal probability of being selected. However, one can see disordered thresholds as category 2 does not have a range along the scale where it is the most likely category to be selected. Threshold disordering suggests that the response scale is not working adequately to order participants with distinct levels of QoL. Category 2 could be combined with either category 3 or 1, but we retained the combination that provided the best PSI. Consequently, we combined the intermediate categories (1 and 2) to generate a revised three-category scale as: 0, “very much”; 1, “a little to moderate amount”; and 2, “not at all.” Following this category reorganization, all thresholds were ordered (Fig. b).

Further psychometric assessment of the CarCGQoL questionnaires revealed several items to display notable DIF and overall the items lacked unidimensionality (first contrast 4.9 eigenvalue units and second contrast 3.6 eigenvalue units, and 3 misfitting items; Table 3). The items from both contrasts (I and II) were deleted, following which PCA of the 30-item CarCGQoL questionnaire demonstrated unidimensionality (i.e., all the items were measuring QoL). However, items continued to misfit so were deleted iteratively, and 22 items remained that all fit the Rasch model well. Furthermore, two items showed notable DIF by mode of questionnaire administration and one item showed notable DIF by SES. Those for whom the CarCGQoL was interviewer-administered found it easier to endorse the item “your child will be able to lead a normal life” (2.15 logits) and the item “your child will be able to take care of you when you are old” (1.81 logits) than those who self-administered the questionnaire. In addition, even if having similar levels of QoL, those who belonged to a lower SES found it easier (and scored higher) to endorse the item “your child will be able to take care of you when you are old” (1.84 logits) than those who belonged to a higher SES. As noted in our Methods section, given the notable DIF demonstrated by these two items, we decided to delete them. Following deletion, 20 items remained in the final questionnaire that all fit the Rasch model well, were free from notable DIF and could adequately discriminate between different levels of caregiver's QoL (PSI, 2.51; person reliability, 0.86). Targeting of the item difficulty to participant's QoL was 0.69 logits. Taken together, these fit parameters indicate that the Rasch-guided item reduction resulted in a 20-item CarCGQoL questionnaire that is valid, reliable, and unidimensional for the assessment of the PCG caregiver's QoL.

The mean QoL of the caregivers of those who were administered different language versions of the CarCGQoL was 0.59 ± 1.54 logits for Hindi, 0.91 ± 1.36 logits for Telugu, and 0.96 ± 2.08 logits for Marathi; this difference was not statistically significant (1-way ANOVA, $P = 0.60$), implying that there was no mean difference in participant's QoL and item difficulties across the languages.

Raw Score to Rasch Measure Conversion

To encourage those who wish to use the CarCGQoL questionnaire and also use the benefits of Rasch analysis in the scoring of the questionnaire (but are unfamiliar with the procedure), we have developed ready-to-use spreadsheets for conversion of raw scores to Rasch-scaled scores for the 20-item questionnaire. These sheets can be obtained by contacting the corresponding author, or can be downloaded from the Journal's Web site (see Supplementary Material). However, it should be borne in mind that such conversions will hold only if the population on which it is being tested is similar to that of the present study.

TABLE 3. Validation of the CarCGQoL Questionnaire

Parameters	Rasch Model, Ideal Values	Versions of CarCGQoL Questionnaire		
		Phase I	Phase II	Phase III, Final
No. of items	—	45	30	20
No. of misfitting items	0	3	5	0
Person separation index	>2.00	3.06	2.56	2.51
Person reliability	>0.80	0.90	0.87	0.86
Mean item location	0	0	0	0
Mean person location	0	0.52	0.67	0.69.00
Principal components analysis (eigenvalue for first and second contrast)	<3.0	4.9 (first contrast) 3.6 (second contrast)	2.9 (first contrast) 2.4 (second contrast)	2.5 (first contrast) 2.0 (second contrast)
No. of items displaying notable DIF, >1 logit	0	8	2	0

DISCUSSION

We have designed, developed, and validated a novel CarCGQoL questionnaire for use with caregivers of children with PCG. It is psychometrically robust, short (at 20 items), and relatively easy to complete. It has excellent psychometric properties: unidimensionality, good reliability, good targeting, and well-functioning rating scale.

There have been very few attempts at investigating the experiences of caregivers of children with PCG. For example, the only study in the literature (from India) assessed the magnitude of caregiver burden in PCG, albeit a restricted aspect of QoL, and this was performed using “off-the-shelf” questionnaires, such as the CBQ.²⁴ However, the authors did not report the psychometric properties of the CBQ in their population before using it. Importantly, this study used raw ordinal scores (based on classical test theory [CTT]) rather than item response theory (IRT) methodology. There are some limitations when generic instruments, such as CBQ, are applied to specific conditions, such as those with PCG. Firstly, these are generic instruments, so they are considered to be less sensitive than disease-specific questionnaires and, secondly, such questionnaires target adults as patients and not as caregivers. Measurement of caregiver QoL using these questionnaires is not ideal because patients and caregivers encounter issues differently. For example, a caregiver may not be affected as much in physical functioning compared to patients, so a questionnaire that emphasizes physical functioning may be minimally relevant to a caregiver's QoL. However, the caregiver may suffer from emotional or social impacts that the questionnaire insufficiently identifies, thus, lowering its applicability.

The high psychometric properties of the CarCGQoL questionnaire can perhaps be explained by two factors. First is the use of FGDs with the caregivers in which they provided their views of the impact of PCG on various aspects of their QoL. This more comprehensive method of measuring caregiver's QoL is advantageous from a measurement perspective, as such a tool can capture the relationship between various aspects of the caregiver's QoL, and specific domains of their child's PCG that may be improved by treatment. The use of FGDs is a way to ensure content validity given the engagement of the caregivers who would not traditionally be viewed as “experts.” Our comprehensive approach consisting of obtaining valuable inputs of the caregiver's through FGDs, and the experts in the conceptual framework and items, thereby emphasizing the dyadic nature of caregiving, differentiates the CarCGQoL questionnaire from other measures. Focus group methodology also is advantageous as it provides empirical information with regard to the type of language that caregivers use when describing their experiences and feelings, such that the items, their wordings, and response categories are of direct

relevance to the population of interest. It now is recommended that the conceptual framework of any newly developed patient-reported outcomes be informed by qualitative studies to ensure that it reflects the perspective and disease experience of the population who will be completing the questionnaires.^{42,43}

Secondly, we used Rasch analysis in the development as well as validation stage of the CarCGQoL questionnaire. This modern psychometric approach enabled us to examine several important properties of the questionnaire; for example, its unidimensionality, that is, the extent to which all items measured the same construct and how well each item measures or “fits” the construct.⁴⁴ Nonetheless, the 45-item CarCGQoL questionnaire lacked unidimensionality as was evidenced by PCA of residuals, thereby violating the fundamental requirement of the Rasch measurement model. Unidimensionality is critical to a questionnaire, because in its absence, the user is unsure of the construct under measurement, so it is mandatory that it is reestablished for an optimally functioning questionnaire. Consequently, 15 items were deleted to restore unidimensionality of the CarCGQoL questionnaire as also few misfitting items. This was followed by deletion of 2 DIF-causing items. Thus, the use of a 20-item CarCGQoL total or summary score is valid with interval level properties that now legitimizes the use of parametric statistics for calculation of change scores⁴⁵; for example, comparison of impact of glaucoma surgery on caregiver's QoL in children with PCG.

Among the desirable features of an optimally functioning questionnaire also include lack of DIF and an ability to discriminate among as many groups of participant's QoL as possible and the higher the number of strata, the better is the discriminative ability of the questionnaire.^{34,46} In addition, a good measure should be able to differentiate persons across the full spectrum of ability in the population. The PSI of the CarCGQoL questionnaire is excellent, indicating that it is reliably able to differentiate among different strata of participant's QoL. Although DIF analyses did not detect any item for most of the sociodemographic variables assessed, we did find two items that showed notable DIF by mode of questionnaire administration and SES. The presence of DIF in the two items suggests that these may respond differently across socioeconomic groups and across modes of questionnaire administration. It is plausible that caregivers in the lower SES and those in whom the questionnaire was interviewer-administered had a desire to give a socially appropriate response to both these items resulting in notable DIF. Interestingly, over half of the participants (58%) in whom the CarCGQoL was interviewer-administered also belonged to the lower SES. Besides the level of DIF that suggested their deletion, these two items were perhaps also not very relevant to caregivers whose children were too young so they could not relate to it resulting in

speculative responses, and problems with DIF will continue to persist given that one of its uses could be as an outcome measure in children undergoing PCG surgery who also are likely to be very young. In addition, our aim was to develop a psychometrically robust questionnaire and given that the presence of DIF violates the requirements of the Rasch model, we decided to delete these two DIF-causing items.

The CarCGQoL questionnaire demonstrated good targeting (0.69 logits) of item severity to the caregiver's QoL. As noted earlier, one of reasons for this good targeting could be related to the generation of item content using FGDs. The original response scale of the CarCGQoL was dysfunctional; however a shortened scale consisting of 3 options functioned well. Smaller numbers of categories (three) are potentially less confusing for the respondents as has been demonstrated by earlier researchers.^{47,48}

Although we set out to develop a caregiver's QoL questionnaire specific to PCG and followed a well-worn path in its development, it appears from the nature of the items that remained in the final CarCGQoL questionnaire that the issues addressed would perhaps be applicable to other chronic pediatric conditions as well (albeit with slight modifications in the framing question to make it condition-specific). However, cross diagnostic validation cannot be assumed and will need to be tested. Nonetheless, if it is shown to possess adequate psychometric properties in other chronic pediatric conditions, then that would offer additional support for its use in these conditions besides PCG, and that would further confirm the results of Rasch analysis in PCG. Results of such cross-diagnostic validation will help optimize its universality across similar chronic pediatric conditions given that not many such instruments are available at present. Furthermore, cross-cultural validation is necessary to suit other cultural or language groups.

Besides acting as a complement to diagnostic information (as mentioned in the Introduction), following completion, the items with lower scores (greater problems), except for item 20, can be used to identify specific issues and be addressed. For example, some caregivers may need one-to-one counseling to allay their fears and concerns regarding their child's PCG. In addition, the information obtained from the CarCGQoL can be used to inform and educate eye care providers about the needs of caregivers of PCG patients. However, an important application of the CarCGQoL could be that it can serve as an outcome measure to evaluate the effectiveness of services so as to improve the caregiver's QoL.

We acknowledge that our study has two important limitations. Firstly, despite our best efforts, our study sample comprised a higher proportion of mothers of children with PCG (both in development and validation phase). Of both parents, however, usually the mother is the primary caregiver in infants. While the over-representation of views of the mothers may have led to a sex bias and influenced the selection of items of final questionnaire, it should not compromise the calibration of the questionnaire. Unlike the calibration of the questionnaire in the traditional test design which is dependent upon the sample, Rasch analysis allows relatively sample-free test calibration.⁴⁹ Secondly, we did not evaluate other important measurement properties, such as test-retest reliability and responsiveness. Further studies will be necessary to evaluate these properties.

In conclusion, the 20-item CarCGQoL questionnaire is a novel measure that has robust psychometric properties using Rasch analysis and is suitable for assessing the QoL of caregivers of children with PCG. The 20-item CarCGQoL questionnaire has potential for use in clinical care as well as an outcome measure in clinical trials of children with PCG. It provides a means for understanding the inter-relationship

between the domains of function of a child with PCG and the QoL as perceived by their caregiver.

Acknowledgments

The authors thank all experts who shared their expertise, time, and opinions, and all participants who volunteered their time to participate in different phases of the study, which helped us to accomplish this research.

Presented in part as a poster at the annual meeting of the Association for Research in Vision and Ophthalmology, Orlando, Florida, United States, May 4–8, 2014.

Supported in part by Hyderabad Eye Research Foundation, Hyderabad, India. The authors alone are responsible for the content and writing of the paper.

Disclosure: **V.K. Gothwal**, None; **S. Bharani**, None; **A.K. Mandal**, None

References

- Mandal AK, Netland PA. Epidemiology and genetics of developmental glaucomas. In: Mandal AK, Netland PA, eds. *The Pediatric Glaucomas*. Philadelphia, PA: Elsevier Butterworth Heinemann; 2006:19–22.
- Franks W, Taylor D. Congenital glaucoma—a preventable cause of blindness. *Arch Dis Child*. 1989;64:649–650.
- Dandona L, Williams JD, Williams BC, Rao GN. Population-based assessment of childhood blindness in southern India. *Arch Ophthalmol*. 1998;116:545–546.
- Francois J. Congenital glaucoma and its inheritance. *Ophthalmologica*. 1972;181:61–73.
- Panicker SG, Reddy AB, Mandal AK, et al. Identification of novel mutations causing familial primary congenital glaucoma in Indian pedigrees. *Invest Ophthalmol Vis Sci*. 2002;43:1358–1366.
- Reddy AB, Panicker SG, Mandal AK, Hasnain SE, Balasubramanian D. Identification of R368H as a predominant CYP1B1 allele causing primary congenital glaucoma in Indian patients. *Invest Ophthalmol Vis Sci*. 2003;44:4200–4203.
- Jaffar MS. *Care of the Infantile Glaucoma Patient*. *Ophthalmology Annual*. New York, NY: Raven Press; 1988:15–37.
- Panicker SG, Mandal AK, Reddy AB, Gothwal VK, Hasnain SE. Correlations of genotype with phenotype in Indian patients with primary congenital glaucoma. *Invest Ophthalmol Vis Sci*. 2004;45:1149–1156.
- Mandal AK, Naduvilath TJ, Jayagandan A. Surgical results of combined trabeculotomy-trabeculectomy for developmental glaucoma. *Ophthalmology*. 1998;105:974–982.
- Mandal AK, Gothwal VK, Nutheti R. Surgical outcome of primary developmental glaucoma: a single surgeon's long-term experience from a tertiary eye care centre in India. *Eye (Lond)*. 2007;21:764–774.
- Broughton WL, Parks MM. An analysis of treatment of congenital glaucoma by goniotomy. *Am J Ophthalmol*. 1981; 91:566–572.
- Richardson KT Jr, Ferguson WJ Jr, Shaffer RN. Long-term functional results in infantile glaucoma. *Trans Am Acad Ophthalmol Otolaryngol*. 1967;71:833–837.
- Biglan AW, Hiles DA. The visual results following infantile glaucoma surgery. *J Pediatr Ophthalmol Strabismus*. 1979;16: 377–381.
- Morgan KS, Black B, Ellis FD, Helveston EM. Treatment of congenital glaucoma. *Am J Ophthalmol*. 1981;92:799–803.
- Elder MJ. Combined trabeculotomy-trabeculectomy compared with primary trabeculectomy for congenital glaucoma. *Br J Ophthalmol*. 1994;78:745–748.

16. Mandal AK, Bhatia PG, Bhaskar A, Nutheti R. Long-term surgical and visual outcomes in Indian children with developmental glaucoma operated on within 6 months of birth. *Ophthalmology*. 2004;111:283-290.
17. McPherson SD Jr, McFarland D. External trabeculotomy for developmental glaucoma. *Ophthalmology*. 1980;87:302-305.
18. Mandal AK, Gothwal VK, Bagga H, Nutheti R, Mansoori T. Outcome of surgery on infants younger than 1 month with congenital glaucoma. *Ophthalmology*. 2003;110:1909-1915.
19. Chow MY, Morrow AM, Cooper Robbins SC, Leask J. Condition-specific quality of life questionnaires for caregivers of children with pediatric conditions: a systematic review. *Qual Life Res*. 2013;22:2183-2200.
20. WHOQOL group. Development of the WHOQOL: rationale and current status. *Int J Mental Health*. 1994;23:24-56.
21. Juniper EF, Guyatt GH, Feeny DH, Ferrie PJ, Griffith LE, Townsend M. Measuring quality of life in the parents of children with asthma. *Qual Life Res*. 1996;5:27-34.
22. Minaya P, Baumstarck K, Berbis J, et al. The CareGiver Oncology Quality of Life questionnaire (CarGOQoL): development and validation of an instrument to measure the quality of life of the caregivers of patients with cancer. *Eur J Cancer*. 2012;48:904-911.
23. The WHOQOL Group. The World Health Organization Quality of Life Assessment (WHOQOL): development and general psychometric properties. *Social Sci Med*. 1998;46:1569.
24. Dada T, Aggarwal A, Bali SJ, Wadhvani M, Tinwala S, Sagar R. Caregiver burden assessment in primary congenital glaucoma. *Eur J Ophthalmol*. 2013;23:324-328.
25. Two R, Verjee-Lorenz A, Clayson D, Dalal M, Grotzinger K, Younossi ZM. A methodology for successfully producing global translations of patient reported outcome measures for use in multiple countries. *Value Health*. 2010;13:128-131.
26. Wackerbarth SB, Streams ME, Smith MK. Capturing the insights of family caregivers: survey item generation with a coupled interview/focus group process. *Qual Health Res*. 2002;12:1141-1154.
27. Streiner DL, Norman GR. *Health Measurement Scales: A Practical Guide to Their Development and Use*, 4th ed. Oxford, UK: Oxford University Press; 2008.
28. O'Donnell AB, Lutfey KE, Marceau LD, McKinlay JB. Using focus groups to improve the validity of cross-national survey research: a study of physician decision making. *Qual Health Res*. 2007;17:971-981.
29. Ferrans CE. Development of a conceptual model of quality of life. *Sch Inq Nurs Pract*. 1996;10:293-304.
30. Wills GB. *Cognitive Interviewing: A Tool for Improving Questionnaire Design*. Thousand Oaks, CA: Sage; 2005.
31. Andrich DA. A rating scale formulation for ordered response categories. *Psychometrika*. 1978;43:361-373.
32. Linacre JM. A user's guide to Winsteps Ministep Rasch-model computer programs: program manual 3.74.0. Available at: www.winsteps.com/a/winsteps-manual-3740.pdf. Accessed September 5, 2014.
33. Massof RW. The measurement of vision disability. *Optom Vis Sci*. 2002;79:516-552.
34. Pesudovs K, Burr JM, Harley C, Elliott DB. The development, assessment, and selection of questionnaires. *Optom Vis Sci*. 2007;84:663-674.
35. Wright BD, Linacre JM, Smith RM, Heinemann AW, Granger CV. FIM measurement properties and Rasch model details. *Scand J Rehabil Med*. 1997;29:267-272.
36. Gothwal VK, Wright TA, Lamoureux EL, Pesudovs K. Activities of Daily Vision Scale: what do the subscales measure? *Invest Ophthalmol Vis Sci*. 2010;51:694-700.
37. Massof RW. An interval-scaled scoring algorithm for visual function questionnaires. *Optom Vis Sci*. 2007;84:689-704.
38. Gothwal VK, Wright TA, Lamoureux EL, Pesudovs K. Visual Activities Questionnaire: assessment of subscale validity for cataract surgery outcomes. *J Cataract Refract Surg*. 2009;35:1961-1969.
39. Wright BD, Douglas GA. *Best Test Design and Self-Tailored Testing. MESA Research Memorandum No 19*. Chicago, IL: Statistical Laboratory, Department of Education, University of Chicago; 1975.
40. Wright BD, Douglas GA. *Rasch Item Analysis by Hand. MESA Research Memorandum No 21*. Chicago, IL: Statistical Laboratory, Department of Education, University of Chicago; 1976.
41. Tennant A, Pallant JFDIF. Matters: a practical approach to test if differential item functioning makes a difference. *Rasch Meas*. 2007;1082-1084.
42. Rothman ML, Beltran P, Cappelleri JC, Lipscomb J, Teschendorf B. Patient-reported outcomes: conceptual issues. *Value Health*. 2007;10(suppl 2):S66-S75.
43. Frank L, Howard K, Jones R, et al. A qualitative assessment of the concept of dependence in Alzheimer's disease. *Am J Alzheimers Dis Other Demen*. 2010;25:239-247.
44. Bond TG, Fox CM. *Applying the Rasch Model: Fundamental Measurement in the Human Sciences*. Londo, UK: Lawrence Erlbaum Associates; 2001.
45. Tennant A, McKenna SP, Hagell P. Application of Rasch analysis in the development and application of quality of life instruments. *Value Health*. 2004;7(suppl 1):S22-S26.
46. Mallinson T. Why measurement matters for measuring patient vision outcomes. *Optom Vis Sci*. 2007;84:675-682.
47. Marella M, Gothwal VK, Pesudovs K, Lamoureux E. Validation of the visual disability questionnaire (VDQ) in India. *Optom Vis Sci*. 2009;86:E826-E835.
48. Gothwal VK, Wright TA, Lamoureux EL, Pesudovs K. Rasch analysis of the quality of life and vision function questionnaire. *Optom Vis Sci*. 2009;86:E836-E844.
49. Wright BD. *Best Test Design: Rasch Measurement*. Chicago, IL: MESA Press; 1979.