Comparing children's and parents' perspectives of health outcome in paediatric hydrocephalus

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This study exampled the properties of a child-completed version of the Hydrocephalus Outcome Questionnaire (cHOQ) and compared these with parental responses to the HOQ (parent version). This was a cross-sectional study in the outpatient clinics at three Canadian paediatric hospitals (Toronto, Vancouver, and Halifax). All cognitively-capable children with previously treated hydrocephalus who were aged between 6 and 19 years were eligible. Parents completed the HOQ and the Health Utilities Index Mark 3; children completed the cHOQ. A total of 273 children participated (146 males, 127 females; mean age 14y 1mo, SD 2y 7mo). Internal consistency of the cHOQ was 0.93 and test–retest reliability was 0.86 (95% confidence interval 0.78–0.92). Mother–child agreement and father–child agreement were 0.57 (0.40–0.68) and 0.62 (0.48–0.73) respectively. Agreement was higher for assessments of physical health, but lower for assessments of cognitive health and social–emotional health. There was greater parent–child agreement for older children. When there was disagreement, it seemed that children tended to rate their health better than their parents did. In older children with hydrocephalus, the cHOQ appears to be a scientifically reliable means of assessing long-term outcome. The differences in child and parent perceptions of health need to be appreciated when conducting outcome studies in this population.

Assessment of health outcome in paediatric medicine is challenging due to the difficulty of directly and meaningfully obtaining responses from the children themselves. Completing a questionnaire that asks patients about their health requires a certain level of understanding and appreciation about the concepts of health and self. The age at which a child develops the skills necessary for this task are debatable. Children's perceptions of health and illness change dramatically as they mature.[1,2] Their developmental conception of illness appears to evolve through several levels of explanations, including pre-logical, concrete, and formal-logical (e.g. physiological).[2,3] These stages represent the increasing ability of the child to differentiate themselves conceptually from their environment. The progression through these stages is thought to occur up until 12 years of age, although there is likely to be great individual variation. This issue is further complicated in paediatric hydrocephalus, in which many children have varying levels of cognitive impairment.

Previous research into outcomes of paediatric hydrocephalus has lacked a reliable and valid measure that is specific to this population. To address this, the current authors developed the Hydrocephalus Outcome Questionnaire (HOQ), a 51-item questionnaire completed by the parents of children with hydrocephalus, which has proven reliability and validity.[4,5] The HOQ provides a score of Overall Health and subscores of Physical Health, Cognitive Health, and Social–Emotional Health. It has been shown that internal consistency (Cronbach's alpha) and test–retest reliability of the HOQ are excellent for the Overall Health score and subscores (all values >0.8).[4]

Using parents as respondents can be viewed in at least two different ways. Their role can be perceived as that of a proxy, in which they attempt to answer a questionnaire from the child's perspective, i.e. answer in the way they think the child would. Alternatively, parents can be used as observers of their children and asked to respond to questions based solely on what they have seen of their children (rather than trying to guess the child's perspective). This latter model – parent as observer – was used for the parent version of the HOQ. Given the wide range of ages and cognitive capabilities of children with hydrocephalus, this seemed to be the most appropriate type of respondent to use.

However, there exists a group of older children with hydrocephalus who have a highly developed perception of their own health. The views of these children are valuable to clinicians, and may well provide unique insight. Therefore, the current authors chose to investigate the feasibility and scientific properties of a child-completed version of the HOQ (cHOQ). The objectives were to: (1) establish the reliability of the cHOQ; (2) determine if there is a minimum age for reliable completion of the cHOQ; and (3) compare child and parental responses to determine whether children perceive themselves to be better, worse, or about the same as their parents, and if there is an child age-effect to this.

Method

Patients with hydrocephalus attending a regularly scheduled neurosurgery outpatient clinic at three paediatric hospitals were eligible to participate. Recruitment started in July 2004 at the Hospital for Sick Children, Toronto and in January 2006 at BC Children's Hospital, Vancouver, and IWK Health Centre, Halifax. Recruitment ended in November 2006. All

children with a history of surgically treated hydrocephalus, which was initially treated at least 6 months earlier, and who were between the ages of 6 and 19 years were considered for this study. A child was deemed eligible if, by parental report, the child had the ability to read English at least at first grade level and attended a regular school, with or without the need for special educational assistance. If the child provided appropriate assent or consent, the child completed the cHOQ (Appendix SI, supplementary material published online).

The cHOQ takes the same 51 items from the parent version of the HOQ, but the items in the Likert scale are presented from the perspective of the child. These items were generated from focus groups and individual interviews that involved parents and children with hydrocephalus.[4] Completion time for the cHOQ is approximately 12 to 18 minutes. Scores for the cHOQ (Overall Health score plus three subscores: Physical, Cognitive, and Social–Emotional Health) are calculated in the same way as for the HOQ (Table SI, supplementary material published online).

Approximately 10 days after the first administration, a second cHOQ was mailed to the child's residence for repeat completion. The completed cHOQ was returned in a stamped, pre-addressed envelope. The required sample size was estimated based on testing the null hypothesis of H_o: $\rho=\rho_o$ against the alternative hypothesis H_1: $\rho>\rho_o$ (where ρ_o is the minimally acceptable reliability coefficient).[6,7] Under the assumptions of $\rho_o=0.70$, alpha=0.05, beta=0.20, and an expected reliability of 0.85, the model predicts a required sample size of 43 participants. To account for non-responders, a repeat questionnaire was sent to the first 125 children seen at the Hospital for Sick Children, Toronto.

Primary caregivers who were willing to participate were asked to complete the standard, parent version of the HOQ and the Health Utilities Index Mark 3 (HUI3).[8] The parent version of the HOQ is a reliable and previously validated measure of health outcome with scores ranging from 0 (worse health) to 1.0 (better health).[4,5] The HUI3 provides a utility score (a preference score) which is also on a metric ranging from 0 to 1, although negative scores (i.e. health states 'worse than death') are possible. The HUI3 assesses the attributes of vision, hearing, speech, ambulation, dexterity, emotion, cognition, and pain.

This project was approved by the local research ethics boards at each centre and informed consent was obtained from all participating families.

STATISTICAL ANALYSIS

Results of the cHOQ assessments were used to calculate internal consistency (Cronbach's alpha) and test–retest reliability (as an intraclass correlation coefficient [ICC] with time as a random factor). Agreement between parent and child responses was calculated as an ICC with rater as a random factor. This was performed separately for mother–child comparisons and father–child comparisons. Floor and ceiling effects were considered important if greater than 15% of respondents endorsed the lowest or highest possible cHOQ score or subscore.[9]

Convergent validity for the cHOQ was tested by assessing Pearson's correlation with HUI3 utility scores. It was hypothesized that there would be a moderate (>0.5) positive correlation. Divergent validity was assessed by comparing cHOQ to child age using Pearson's correlation: it was hypothesized that there would be no significant correlation ($p<0.05$). Known-groups validity was assessed based on the known association between long-term outcome and prolonged hospital stay related to hydrocephalus complications.[10] It was hypothesized that there would be lower cHOQ scores for children who had required at least 14 days of cumulative in-patient stay over their lives.

All statistical analyses were performed using SPSS (version 13.0).

Results

A total of 273 children (146 males, 127 females) participated and completed the cHOQ during their clinic visit (Toronto $n=210$, Vancouver $n=39$, Halifax $n=24$). A total of 324 other children with hydrocephalus did not meet the inclusion criteria for this study, because they were either not cognitively capable of completing the cHOQ or refused to do so (Toronto $n=274$, Vancouver $n=34$, Halifax $n=16$). Mean parental HOQ Overall Health score was significantly higher for the 273 children who completed the cHOQ compared with the 324 children who did not (0.71 vs 0.61, $p<0.001$, independent samples *t*-test). Mean age of the 273 participating children was 14 years, 1 month, which was significantly older than the children who did not participate (mean age 10y 1mo, $p<0.001$). Other characteristics of participating children are shown in Table I and Figure 1.

RELIABILITY

Seventy-six of 125 children (61%) completed and returned a repeat questionnaire. The following variables were not significantly different between the children who responded to the repeat cHOQ and those who did not: current age, age at first treatment, initial cHOQ Overall Health score and subscores (all $p>0.50$, independent samples *t*-test). All reliability coefficients for the cHOQ were greater than 0.70 (Table II).

Table I: Participant characteristics

Variable	
Current age, mean (SD)	14y 1mo (2y 7mo)
Age at first surgery, mean (SD) mo	23 (44.9)
Males/females	146/127
Aetiology of hydrocephalus, *n* (%)	
Myelomeningocele	93 (34.1)
IVH of prematurity	30 (11.0)
Aqueductal stenosis	28 (10.3)
Post-infectious	11 (4.0)
Congenital communicating	7 (2.6)
Intracranial cyst	22 (8.1)
Others	82 (30.0)
Total cumulative number of days in hospital related to hydrocephalus, mean (SD)	32.3 (31.8)
cHOQ Overall Health score, mean (SD)	0.78 (0.14)
cHOQ Physical Health score, mean (SD)	0.82 (0.16)
cHOQ Cognitive Health score, mean (SD)	0.71 (0.20)
cHOQ Social–Emotional Health score, mean (SD)	0.79 (0.15)
HUI3 utility score, mean (SD)	0.71 (0.27)

IVH, intraventricular hemorrhage; cHOQ, child-completed Hydrocephalus Outcome Questionnaire; HUI3, Health Utilities Index Mark 3.

Floor and ceiling effects did not appear to be a concern. No respondent endorsed the lowest possible score and no more than 6.2% endorsed the highest possible score for the cHOQ Overall Health score and for all subscores.

VALIDITY

There was a moderate positive correlation between cHOQ Overall Health score and HUI3 utility score (Pearson[2] correlation 0.60, $p<0.001$). This confirmed the authors' convergent validity hypothesis. There was no significant correlation between cHOQ Overall Health score and child age (Pearson's correlation 0.01, $p=0.86$), confirming the divergent validity hypothesis. Mean cHOQ Overall Health score was significantly lower for children who had spent 14 or more cumulative days in hospital for hydrocephalus treatment than for those who had spent fewer than 14 days (0.75 vs 0.81, $p=0.001$, *t*-test). This was also the case for all cHOQ subscores, thus confirming the known-groups validity hypothesis.

PARENT–CHILD AGREEMENT

A total of 230 mothers and 113 fathers of the children participating completed the HOQ. Interrater agreements between mother–child and father–child scores are listed in Table SI. Agreements for Physical Health scores were high (>0.74), while all other scores had agreements ranging between 0.41 and 0.51. Scatterplots of the relationship between mother–child and father–child HOQ scores demonstrate that most disagreements involved the child rating their own health as better than their parent's rating (Fig. 2).

ANALYSIS BY AGE GROUP

Test–retest reliability, internal consistency, and mother–child agreement for each age quintile range (<11.9, 11.9 to <13.4, 13.4 to <14.7, 14.7 to <16.7, and ≥16.7y) were calculated (Table SIIa–c, supplementary material published online). Father–child comparisons were not examined as there were too few father responses. Although the reliability estimates appeared to be acceptable across all age groups, mother–child agreement was noticeably lower in the youngest and, to a lesser extent, the second youngest age group.

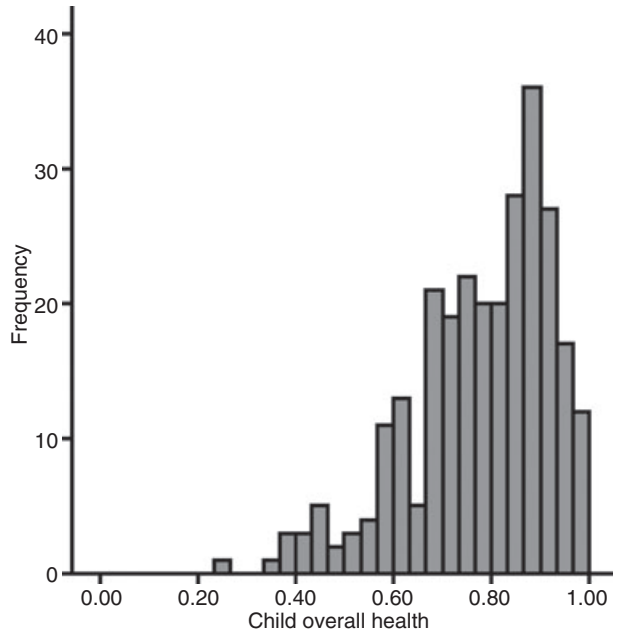

Figure 1: *Distribution of Overall Health scores from the child-completed version of the Hydrocephalus Outcome Questionnaire.*

Discussion

This study assessed the reliability of a child-completed version of the HOQ in a group of older children with hydrocephalus and compared their perceptions of their health with those of their parents.

RELIABILITY AND VALIDITY OF THE CHOQ

The cHOQ had promising reliability in the current sample of children. Analysis by age-group was particularly revealing in showing that these reliability coefficients were consistently good across all age quintiles. By most standards, the level of cHOQ reliability demonstrated in this study would be considered more than adequate for research use (internal consistency ranging from 0.82 to 0.93 and test–retest reliability ranging from 0.77 to 0.90). In general terms, this translates to the children responding to each of the 51 questions in a consistent manner (consistent both *within* each questionnaire administration and *between* two separate administrations of the questionnaire). It should be noted that the sample recruited for this study was a select group of children with hydrocephalus who were older and had generally attained better health outcomes. However, this was not a negligibly small group, as it represented about 46% of those children with hydrocephalus seen in the participating institutions. Among those children aged 10 years or older, 66% (263 of 401) participated. Therefore, there was a sizable majority of older children to whom the cHOQ could be administered with scientifically meaningful results.

In previous publications the current authors helped to establish the construct validity of the HOQ.[4,5] Good correlation was documented between HOQ scores and various other independent measures of health, including the HUI,[8] the Strengths and Difficulties Questionnaire,[11] the Wide Range Achievement Test,[12] and the Functional Independence Measure for Children.[13] In the current study, there was good correlation (0.60) between cHOQ scores and HUI3 utility scores (another validated measure of general health status). The study's divergent validity and known-groups validity hypotheses were also confirmed. The cHOQ demonstrated no worrisome ceiling effect, unlike the generic HUI

Table II: Reliability estimates for the child-completed Hydrocephalus Outcome Questionnaire (cHOQ)

	Internal consistency (Cronbach's alpha)	*Test–retest reliability ICC (95% CI)*
cHOQ Overall Health score	0.93	0.86 (0.78–0.92)
cHOQ Physical Health score	0.82	0.90 (0.84–0.94)
cHOQ Cognitive Health score	0.88	0.77 (0.65–0.85)
cHOQ Social–Emotional Health score	0.85	0.79 (0.68–0.86)

ICC, intraclass correlation coefficient; CI, confidence interval.

utility score for which 21% of the sample endorsed the highest score. This reinforces confidence in the ability of the cHOQ to measure health outcome in a more accurate way.

PARENT–CHILD COMPARISON

In other disease conditions, correlations between parent and child perceptions of health have been reported as no better than moderate, with slightly better correlations for more observable aspects of health (like physical functioning).[14–23] In some reports, children tended to rate themselves worse off than perceived by the parents.[17,23] It has also been reported that parents may underestimate their children's pain and some of the negative emotions associated with illness.[24,25] However, in other studies, parents tended to rate their child's well-being as lower than reported by the child.[15,18,19] Given the different ways in which children and adults might conceptualize health and illness, it has been argued that we should not expect there to be substantial correlations between their ratings of health.[26]

The current study is, to the authors' knowledge, the first to report parent–child comparison in paediatric hydrocephalus. Overall agreement between parent and child dyads was reasonably good (ICC=0.57, 95% CI 0.40–0.68; and ICC=0.62, 95% CI 0.48–0.73), but there was wide variation depending on what aspect of health was being measured. Physical Health scores (an easily *observable* aspect of health) displayed high agreement (ICC=0.74, 95% CI 0.65–0.80; and

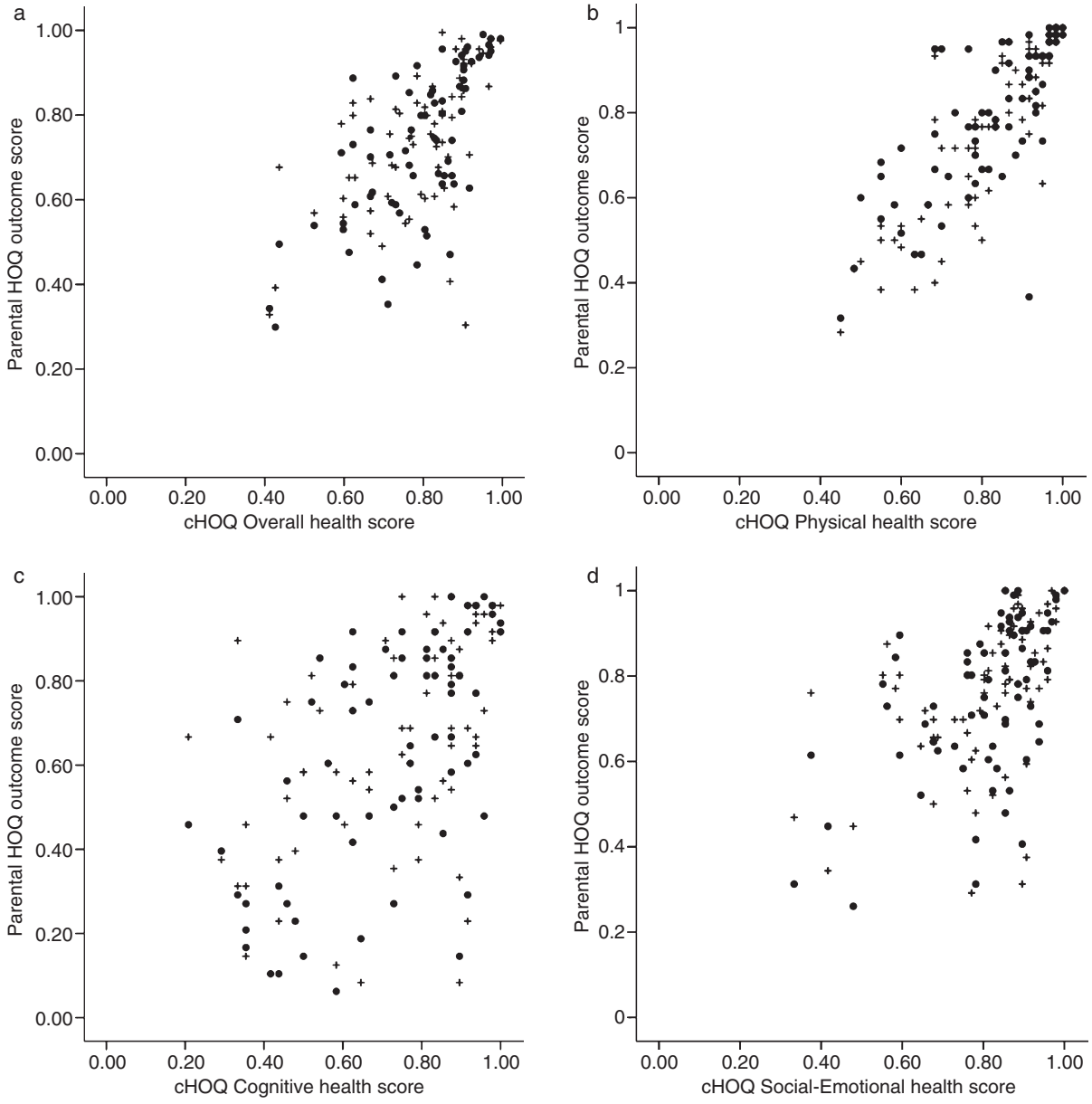

Figure 2. *Relationship between parental Hydrocephalus Outcome Questionnaire (HOQ) scores and child-completed HOQ (cHOQ) scores for: (a) Overall Health scores; (b) Physical Health scores; (c) Cognitive Health scores; (d) Social–Emotional Health scores. Mother–child pairs are marked as dots (•) and father–child pairs as crosses (+).*

ICC=0.75, 95% CI 0.65–0.83), while this was not so much the case for Cognitive and Social–Emotional Health scores (both clearly less observable aspects of health). The bulk of disagreements relate to children rating their status as *better* than their parents. It is not clear how this type of disagreement should be interpreted. Parents, for example, might be influenced by having a higher level of expectation for their child's level of cognitive or social functioning or by their concerns over their child's future performance and independence, rather than just their current status.

There did appear to be an age-related effect in parent–child agreement. It was only in the second quintile and older that agreements were observed at >0.5 with some consistency. This represents those children aged 11 years 10 months and older. Interestingly, this is concordant with the perceived evolution of a child's conceptualization of illness through developmental stages which usually mature at about age 12 years.[2,3] Therefore, it is possible that at this age the child's concept of illness better matches that of the parent's, hence the higher agreement in measures of health outcome.

LIMITATIONS

One limitation of the test–retest reliability measurement is the 61% response rate to repeat administration of the cHOQ. If only the more motivated individuals contributed to the test–retest reliability coefficient, this could have provided a somewhat artificially high estimate. The current analysis of reliability and agreement among the quintiles of child age involved a somewhat limited sample size within each group. Therefore, the precision of some of the estimates was limited and had large confidence intervals. This was especially the case for the test–retest reliability assessment, which assessed fewer than 20 children per quintile group. In assessing convergent validity, this study only compared the cHOQ with the HUI3. In future work, the current authors hope to expand comparison with other child-completed measures of outcome.

Conclusions

In many older children with hydrocephalus, the cHOQ appears to be a scientifically reliable means of assessing long-term outcome. Perceptions of physical health appear to be similar for parents and children, but perceptions differ for cognitive and social–emotional health. In general, it appears that children provide a more optimistic view of their own status than their parents do. Parent–child agreement was especially poor for the youngest children. Future studies assessing hydrocephalus outcomes should attempt to include the child's perspective whenever possible, as it might provide unique insight.

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Supplementary material

The following supplementary material is available for this article online:

Appendix S1: The child-completed Hydrocephalus Outcome Questionnaire (cHOQ) with instructions for administration and scoring.

Table SI: Parent–child agreement for the Hydrocephalus Outcome Questionnaire (HOQ) and child-completed HOQ (cHOQ).

Table SIIa: Parent–child agreement for the Hydrocephalus Outcome Questionnaire (HOQ) and child-completed HOQ (cHOQ).

Table SIIb: Internal consistency estimates for quintiles of age.

Table SIIc: Mother–child agreement estimates for quintiles of age.

This material is available as part of the online article from http://www.blackwell-synergy.com/doi/abs/10.1111/j.1469-8749.2008.03037.x (this will link you to the article abstract)

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