

## Health-related quality of life in pediatric Chiari Type I malformation: the Chiari Health Index for Pediatrics

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**OBJECTIVE** The purpose of this study was to design and validate a patient-reported health-related quality of life (HRQOL) instrument for pediatric Chiari Type I malformation (CM-I), the Chiari Health Index for Pediatrics (CHIP).

**METHODS** The CHIP has 45 items with 4 components making up 2 domain scores, physical (pain frequency, pain severity, nonpain symptoms) and psychosocial; physical and psychosocial scores are combined to create an overall HRQOL score. Increasing scores (0 to 1) represent increasing HRQOL. Fifty-five patients with CM-I (mean age  $12 \pm 4$  years, 53% male) were enrolled and completed the CHIP and Health Utilities Index Mark 3 (HUI3). Twenty-five healthy controls (mean age  $11.9 \pm 4$  years, 40% male) also completed the CHIP. CHIP scores were compared between these groups via the Mann-Whitney U-test. For CHIP discriminative function, subscore versus presence of CM-I was compared via receiver operating characteristic curve analysis. CHIP scores in the CM-I group were stratified by symptomatology (asymptomatic, headaches, and paresthesias) and compared via Kruskal-Wallis test with Mann-Whitney U-test with Bonferroni correction ( $p < 0.0167$ ). CHIP was compared with HUI3 (Health Utilities Index Mark 3) via univariate and multivariate linear regression.

**RESULTS** CHIP physical and psychosocial subscores were, respectively, 24% and 18% lower in CM-I patients than in controls ( $p < 0.001$ ); the overall HRQOL score was 23% lower as well ( $p < 0.001$ ). The area under the curve (AUC) for CHIP physical subscore versus presence of CM-I was 0.809. CHIP physical subscore varied significantly with symptomatology ( $p = 0.001$ ) and HUI3 pain-related quality of life ( $R^2 = 0.311$ ,  $p < 0.001$ ). The AUC for CHIP psychosocial subscore versus presence of CM-I was 0.754. CHIP psychosocial subscore varied significantly with HUI3 cognitive- ( $R^2 = 0.324$ ,  $p < 0.001$ ) and emotion-related ( $R^2 = 0.155$ ,  $p = 0.003$ ) quality of life. The AUC for CHIP HRQOL versus presence of CM-I was 0.820. Overall CHIP HRQOL score varied significantly with symptomatology ( $p = 0.001$ ) and HUI3 multiattribute composite HRQOL score ( $R^2 = 0.440$ ,  $p < 0.001$ ).

**CONCLUSIONS** The CHIP is a patient-reported, CM-I-specific HRQOL instrument, with construct validity in assessing pain-, cognitive-, and emotion-related quality of life, as well as symptomatic features unique to CM-I. It holds promise as a discriminative HRQOL index in CM-I outcomes assessment.

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**KEY WORDS** Chiari Type I malformation; quality of life; Chiari Health Index for Pediatrics

**I**N much of the neurosurgical literature on pediatric Chiari Type I malformation (CM-I), the focal point of surgical outcomes research has been on technical success and reduction in preoperative symptoms.<sup>4,7,17,27,29,38,39,41</sup> However, there are limited reports on patients' experiences via assessing changes in health-related quality of life (HRQOL). HRQOL is important, as pain can have a critical impact on a child's overall well-being, including physical, social, emotional, and cognitive functioning.<sup>10,34,37</sup> These aspects of health can be difficult to measure in a

reproducible and valid manner, particularly in children.<sup>10,42</sup> To date there are currently no standardized disease-specific quality of life measurement instruments designed and validated for use for pediatric patients with CM-I.

Currently, numerous HRQOL and pain scales exist. The SF-36<sup>3</sup> or SF-12<sup>11</sup> both measure physical and mental health, and the Health Utilities Index<sup>8</sup> measures comprehensive health status. The EQ-5D<sup>18</sup> is a patient self-reported health and pain scale. All of these instruments have been validated and are frequently used for patients 18 years of age

**ABBREVIATIONS** AUC = area under the curve; CCOS = Chicago Chiari Outcomes Scale; CHIP = Chiari Health Index for Pediatrics; CM-I = Chiari Type I; HOQ = Hydrocephalus Outcomes Questionnaire; HRQOL = health-related quality of life; HUI3 = Health Utilities Index Mark 3; PedsQL 4.0 = Pediatric Quality of Life Inventory 4.0; ROC = receiver operating characteristic; SF-10 = SF-10 Health Survey for Children.

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and older. Pain scales such as the Neck Disability Index<sup>43</sup> and visual analog scale<sup>5</sup> are commonly used to measure pain in a variety of adult and childhood diseases but are not specific to CM-I. Most of these instruments have only been validated in adult patient populations and are not adaptable, or have not yet been found to be reliable, in the pediatric patient population.

The SF-10 Health Survey for Children (SF-10)<sup>40</sup> is a parent-completed survey that contains 10 questions adapted from the Child Health Questionnaire. The SF-10 provides coverage across a wide range of domains, and is scored to produce physical and psychosocial health summary measures. The Health Utilities Index Mark 3 (HUI3),<sup>9</sup> a multiattribute (generic) status classification system, has previously been validated in the pediatric population for numerous disease states and is both reliable and generalizable in pediatric patients with illness. The HUI3 assesses domains including vision, hearing, speech, ambulation, dexterity, emotion, cognition, and pain. The Pediatric Quality of Life Inventory 4.0 (PedsQL 4.0)<sup>42</sup> is another multidimensional quality of life instrument for children, assessing physical health, psychosocial health, emotional functioning, social functioning, and school functioning.

However, generic HRQOL instruments suffer from an important shortcoming, namely that they are designed to be applicable to a wide variety of populations with many different underlying conditions. As a result, they may not be sufficiently sensitive for capturing meaningful differences among patients with a specific condition or chronic symptoms. Research specifically evaluating HRQOL in pediatric CM-I patients is also limited. One scale, The Chicago Chiari Outcomes Scale (CCOS)<sup>1,44</sup> was developed to assess pain, nonpain symptoms, functionality, and complications in patients with Chiari malformation. However, the shortcoming of this particular instrument is that it does not involve patient input, but rather relies on the clinician to assess pain and function. The patient-reported aspect of HRQOL is important, as children with chronic diseases report a significantly lower HRQOL than healthy children of the same age,<sup>6</sup> although limited data exist.

While several studies have been conducted to assess the reliability and generalizability of HRQOL instruments used in adult CM-I patients,<sup>12,30,31</sup> very few instruments appropriately evaluate the pediatric population. For this reason we have developed the Chiari Health Index for Pediatrics (CHIP), a multidomain 45-item instrument that provides a quantitative score in the domains of somatic complaints and psychosocial function. The purpose of this study is to develop and validate a reliable and simple questionnaire for pediatric patients with CM-I as a quantitative indicator of HRQOL for clinical and research purposes.

In this article, we first evaluate HRQOL in patients with CM-I using an existing generic classification system, the HUI3,<sup>2,20,33</sup> which has previously been validated in healthy children and numerous disease states, but not yet CM-I. We then compare HUI3 to the intentionally disease-specific CHIP instrument. We report on the development and validation of CHIP, discuss its psychometric properties, and provide normative data for this population. We describe in particular the feasibility, internal consistency, and construct validity of CHIP.

## Methods

### Subjects and Settings

All procedures in this study were carried out following institutional review board approval. Subjects were children ages 5–18 years and their caregivers, with 80 participants enrolled overall. Subjects were children presenting at the pediatric neurosurgery clinic at the Children's Hospital at Vanderbilt with a radiological diagnosis of CM-I. Surveys were administered to patients during their clinic visit. Of the children who enrolled into the study, 55 had never undergone surgical decompression. This current report concerns these 55 patients.

The mean  $\pm$  SD age of the 29 boys (53%) and 26 girls (47%) was  $12 \pm 4.0$  years (range 5–17 years) (Table 1). The sample was heterogeneous with respect to race/ethnicity, with 49 white non-Hispanic/Latino (89%), 1 white, Hispanic/Latino (2%), and 5 black/African American, non-Hispanic/Latino patients (9.1%).

For each patient, clinical and radiographic data were obtained from standardized physician and radiographic reports in the electronic medical record. With regard to the symptomatology reported in the examining neurosurgeon's clinical note, patients were assigned to 1 of 3 categories: asymptomatic ( $n = 10$ , 18%), headaches only ( $n = 37$ , 67%), or extremity sensory complaints (i.e., pain, numbness, paresthesias;  $n = 8$ , 15%). With regard to radiological data, patients were divided into 1 of 2 categories: syrinx ( $n = 12$ , 22%) or no syrinx ( $n = 43$ , 78%).

### Surveys

#### Development of CHIP

*Previous HRQOL Pilot Study.* In an effort to identify an existing HRQOL instrument for use in the CM-I patient population we conducted a prospective pilot study using the SF-10. The survey was complemented with demographic and clinical data. Comparisons were made between the SF-10 normative data scores and the scores from our pediatric CM-I patients. Eighty patients were seen dur-

**TABLE 1. Summary of demographic and clinical characteristics of the 55 study participants with CM-I\***

Characteristic	Value
Age in yrs, mean $\pm$ SD	12 $\pm$ 4.0
Male sex	29 (53)
Race/ethnicity	
White, non-Hispanic/Latino	49 (89)
White, Hispanic/Latino	1 (2)
Black/African American	5 (9)
Symptoms	
Asymptomatic	10 (18)
Headaches only	37 (67)
Paresthesias	8 (15)
Radiologic	
No syrinx	43 (78)
Syrinx	12 (22)

\* All values are n (%) except where indicated.

ing the study period. Fifty-four patients (68%) were found to have confirmed CM-I and completed the surveys. We tested for normality and found that our cohort followed a normal distribution; therefore, parametric statistics were used in our analysis. The mean age was  $10.29 \pm 3.69$  years. The average score for physical health in our CM-I cohort was 37.07 compared with national averages of 52.43 ( $p < 0.0005$ ). Our cohort scored 49.47 on psychosocial health compared with the national average of 52.81 ( $p < 0.0005$ ). The CM-I cohort scored in the 5th and 45th percentiles for physical and psychosocial health, respectively. We compared the scores to patient-reported symptoms but were unable to identify correlation for either physical or psychosocial health. Although the SF-10 was able to evaluate physical and psychosocial health, the instrument was not able to address chronic symptoms or pain related to CM-I diagnosis, and, therefore, was not an appropriate measure of quality of life in this patient population (results presented here have not been previously published). These results informed the development of a pediatric CM-I-specific HRQOL instrument, the focus of this study.

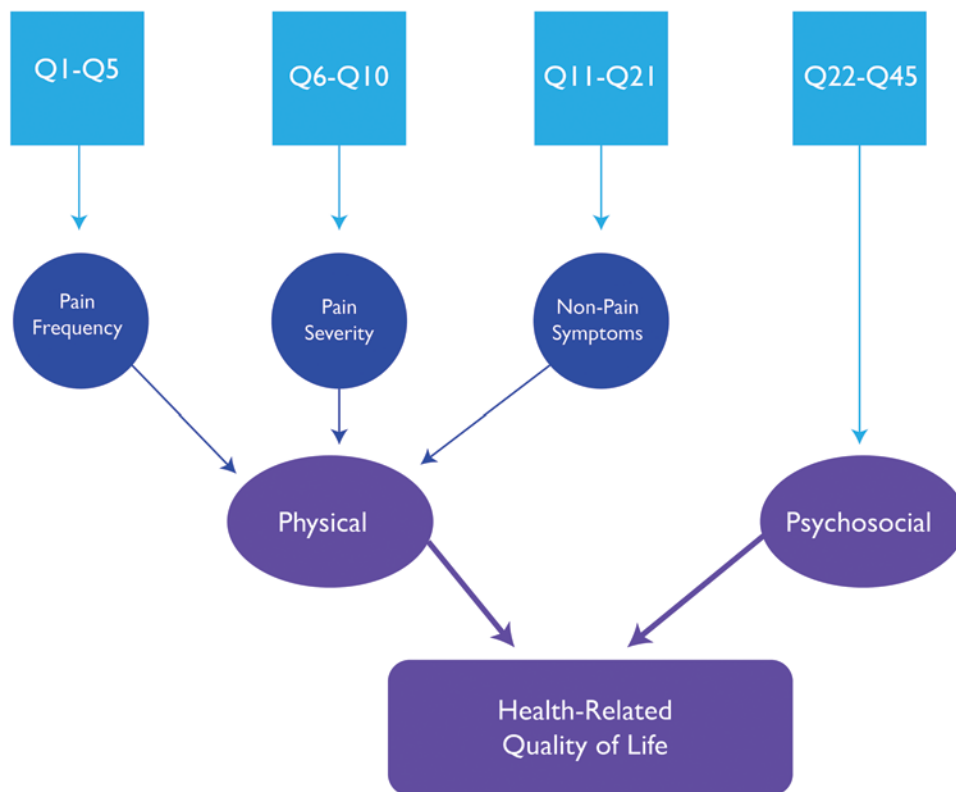
*Item Generation and Development.* When determining how and what a CM-I-specific HRQOL instrument should measure, we first determined that the HRQOL instrument needed to meet the following criteria: 1) it should require patient involvement in answering the questions, 2) it should be age appropriate, 3) it should be thorough yet

concise so that it can be completed in a short time period in the privacy of a patient room, and 4) it should help inform clinical decision making regarding treatment and management of these patients. Taking into consideration these criteria we developed the CHIP.

We identified several HRQOL instruments that had previously been validated and found to be reliable including the HUI3,<sup>9</sup> Hydrocephalus Outcomes Questionnaire (HOQ),<sup>24</sup> PedsQL 4.0,<sup>42</sup> and the SF-10.<sup>41</sup> We then evaluated the questions used in those instruments as a starting point in the development of the CHIP. An initial 50-item instrument was created with 4 components making up 2 domain scores: physical (pain frequency, pain severity, nonpain symptoms) and psychosocial. In addition there was an overall HRQOL score, a weighted average of the physical and psychosocial subscores.

We then tested our items against the 12 threats of content validity (ambiguity, leading questions, double barreled, reverse coding, negative wording, double negatives, jargon, colloquialisms, acronyms, prestige bias, social responsibility bias, and acquiescence bias). We found 2 of our questions to be categorized under “negative wording,” and they were reworded accordingly.

*Factor Analysis.* We then conducted factor analysis by testing each item against its corresponding factor (Fig. 1). Of the 50 items tested, we identified 5 items whose variance was below the a priori cutoff of 0.50, and these were



**FIG. 1.** The CHIP construct matrix showing how each question, component, and domain constitutes overall HRQOL. The top row of blocks represents the question groupings. Blocks Q1–Q5, Q6–Q10, and Q11–Q21 represent the pain frequency, pain severity, and nonpain symptom components, respectively, which further define the physical domain. Block Q22–Q45 represents the psychosocial component, which further defines the psychosocial domain. Both domains are combined to yield the overall HRQOL score. Figure is available in color online only.

subsequently removed from the inventory. All other items had a variance of 0.52–0.99, confirming that each item was appropriately correlated with the factor assigned. We then tested for the significance of factors in our model. No common factors were identified, and the number of factors in our model was found to be sufficient ( $p < 0.0001$ ). Additionally, the total variance explained by the factors was 80.3, and the Tucker-Lewis reliability coefficient was found to be 0.68 ( $> 0.60$  was used as acceptability criteria<sup>36</sup>).

## CHIP

The final CHIP is a single 45-item form, with no variation in question content across age groups. (The CHIP instrument is available as Supplemental Material accessible through a link provided under Online-Only Content in the endmatter). The CHIP can be completed by a parent only, a child only, or a parent and child together. A 5-point response scale is used: (4 = never; 3 = almost never; 2 = sometimes; 1 = often; 0 = almost always). Within the physical symptoms domain, 5 questions regarding pain severity are alternatively scaled as follows: (4 = no pain; 3 = mild; 2 = uncomfortable; 1 = intense; 0 = severe). Scoring information is provided to facilitate clinician and researcher use of this instrument; for each component, the scores for each item are added, and the sum is divided by the product of 4 times the number of answers completed (Fig. 2).

In this way, the score is corrected for missing values and scaled to a range of scores between 0 and 1. An increasing score represents increasing HRQOL for that component. The number of items pertaining to each component is as follows: pain frequency 5, pain severity 5, nonpain symptoms 11, and psychosocial 24. The physical domain score is computed by taking the weighted average of its components: (pain frequency  $\times 3$  + pain severity  $\times 1.5$  + nonpain symptoms)/5.5. The overall HRQOL score is computed by (physical subscore  $\times 3$  + psychosocial subscore)/4. These weightings were determined by 1) comparing the relative contributions of each factor to the overall variance of the instrument and 2) the correlation ( $R^2$ ) between each factor and the HUI3 multiattribute composite HRQOL score.

## HUI3

The HUI3, a multiattribute (generic) status classification system, has previously been validated in the pediatric population for healthy children and also numerous disease states, and is both reliable and generalizable in pediatric patients with acute or chronic illness.<sup>2,8,9,20,33</sup> It is comprised of 8 attributes: vision, hearing, speech, ambulation, dexterity, emotion, cognition, and pain, each with 5 or 6 discriminative levels of ability or disability. These can be expressed as single attributes as well as a multiattribute composite score, which takes into account each factor's effect on HRQOL. The equation for this composite score is: multiplicative score =  $1.371 \times ([\text{multiattribute HUI3 factor}_1] \times \dots \times [\text{multiattribute HUI3 factor}_8]) - 0.371$ .<sup>9</sup> For this study, given that HUI3 has been previously validated in children, this index was used to 1) compare the CM-I population to healthy children, in general, as well as 2) provide a reference index for validating the psychometric properties of CHIP.

$$\text{Score} = \frac{\sum_i^k \text{Item score } (i)}{4 \times (\# \text{ Q's answered})}$$

**FIG. 2.** The equation showing how the CHIP instrument is scored. For each component (i.e., pain frequency, pain severity, nonpain symptoms, psychosocial), the scores for each item ( $i$  through  $k$ ) are added, and the sum is divided by the product of 4 times the number of answers completed (Fig. 1). In this way, the score is corrected for missing values and scaled to a range of scores between 0 and 1. Note: the physical domain score is computed by taking the weighted average of its components: (pain frequency  $\times 3$  + pain severity  $\times 1.5$  + nonpain symptoms)/5.5. The overall HRQOL score is computed by (physical subscore  $\times 3$  + psychosocial subscore)/4.

## Procedure

Inclusion criteria were child age 5 to  $< 18$  years and parent and child English-speaking. The participants were a convenience sample based on consecutive patients presenting at the pediatric neurosurgery clinic. Subjects were identified by selecting for patients being seen for a CM-I consultation, as documented in the clinic appointment scheduling system. After being seen by their neurosurgeon, a member of the research team approached patients and families about potential participation. Once adequately informed of the study and agreement to participate was given, written parental informed consent and child assent, as needed, were each obtained. Patients and/or patients' guardian were asked to complete the CHIP and HUI3 while still in the clinic. The research team member was on standby to collect the instrument once it was completed.

In general, the survey was completed by the parent/guardian alone for young children ( $n = 6$ , mean age =  $9.3 \pm 3.2$  years). For most children, the survey was completed by the parent/guardian in consultation with the child ( $n = 36$ , mean age =  $12 \pm 4$  years). Older children generally completed the survey independently ( $n = 13$ , mean age =  $13.2 \pm 4.1$  years). For children less than 12 years of age, 84% of surveys were completed by the parent/guardian entirely ( $n = 9$ ) or with the help of the parent/guardian ( $n = 16$ ). Overall 86 patients and their caregivers were approached for consent, with 6 declining participation. The main reasons for declining were lack of interest in participating in research and perceiving no direct benefit from the research. Overall, 55 of these patients met the inclusion criteria for no prior CM-I decompression. The estimated time to complete the CHIP was 5 minutes; however, as patients and families were completing surveys during their clinic visit, we allotted 15 minutes.

## Statistical Analysis

### HUI3 HRQOL Data Analysis

All statistical analysis was performed in SPSS Statistics 21 (IBM). Continuous variables were described as mean  $\pm$  SD, and categorical variables were described as number (percentage). Mean HUI3 subscores were compared via Kruskal-Wallis one-way ANOVA with post hoc Mann-Whitney U-test with Bonferroni correction ( $p < 0.0167$ ) for CM-I patients in the following groups: asymptomatic, headaches only, and paresthesias. Normative single-attri-



bute HUI3 values in healthy controls in the childhood age range (ages 5–12, originally published by Pogany et al.) were obtained.<sup>33</sup> In that paper, controls were selected at random across all 10 provinces in Canada, there was a large sample size for each single-attribute HUI3 data point ( $n = 2404$ ), and SD was reported; therefore, we assumed a normal distribution.

We wanted to perform a statistical comparison of single-attribute HUI3 data in the CM-I population in our study to the healthy children in that study. However, because the data in our center were not normally distributed, we were not able to perform a standard, direct parametric comparison using mean,  $n$ , and SD alone. Therefore, we elected to impute data for healthy patients with the intention of performing a nonparametric test of means. Microsoft Excel 2011 was used to generate a normal distribution curve for each single-attribute HUI3 component from the Pogany et al. population. For each of 9 columns representing the 8 HUI3 components plus the mean of the components, 2404 rows were each populated with a random value that would statistically fall within the normal distribution of the sample, using the following formula in each cell: “=NORMINV(RAND(), [single-attribute HUI3 factor mean], [single-attribute HUI3 factor SD]).”

Data obtained in this way were imported into SPSS; the mean and SD for each single-attribute HUI3 factor were calculated to confirm an equivalent distribution as the prior paper. These measures were identical to that reported by Pogany et al. in all cases. Therefore with this comparable artificial dataset, we then compared respective healthy and CM-I single-attribute HUI3 factor scores using the Mann-Whitney U-test, a test for nonparametric data. In addition, a composite HUI3 score, the multiplicative multiattribute score, was calculated for the CM-I population and stratified by symptomatology and compared using Kruskal-Wallis ANOVA with post hoc Mann-Whitney U-test with Bonferroni correction ( $p < 0.0167$ ).

### CHIP Item-Level Analysis

For each item, the distribution of responses and percentage of missing responses were calculated. The Shapiro-Wilk test was performed to determine the normality of item responses, with  $p > 0.05$  indicating a normal distribution (null hypothesis).

### CHIP Scale-Level Analysis

The mean score and range of scores for each CHIP component were determined. The percentage of scores occurring at each extreme of the possible scoring range of 0 or 1, i.e., the ceiling effect (percentage of respondents at highest possible score) and floor effects (percentage of respondents at lowest possible score), respectively, were calculated. For each CHIP component, feasibility (percentage of surveys without any missing responses) was calculated. For each CHIP component, internal consistency was determined by calculating Cronbach's alpha, defining acceptable consistency as  $0.90 \geq \alpha > 0.70$  a priori. In post hoc analysis, test-retest reliability was assessed using a bivariate correlation, defining acceptable reliability as Pearson's  $r > 0.75$  a priori; patients included

in this analysis were all those who had 2 or more survey administrations within 180 days, without a surgical intervention in the interval.

### CHIP Construct Validity

Construct validity was determined via the known-groups technique.<sup>15</sup> A control sample of healthy children ( $n = 25$ , age =  $11.9 \pm 4.0$  years, 40% male) was recruited from members of the Vanderbilt community who volunteered to participate on behalf of their children ( $< 18$  years of age). All children were reported as being free from disease by their parents/guardians, and the CHIP was completed. First, mean CHIP component scores in CM-I patients were compared with mean scores in age-matched healthy controls using the Mann-Whitney U-test. Receiver operating characteristic (ROC) curve analysis was used to measure the discriminative function of CHIP, using CHIP component scores as independent variables and presence of CM-I as the dependent variable. Acceptable discriminative function was defined as area under the curve (AUC)  $> 0.75$ , a priori. Second, mean CHIP reference scores were compared across groups of patients of known differing severity of disease. That is, the CHIP component subscores were compared via Kruskal-Wallis ANOVA with post hoc Mann-Whitney U-test with Bonferroni correction ( $p < 0.0167$ ) for patients in the following groups: asymptomatic, headaches only, and paresthesias. Additionally, CHIP scores were compared with corresponding HUI3 scores within similar domains via univariate linear regression. The following comparisons were made: CHIP physical symptoms versus HUI3 pain and HUI3 dexterity, CHIP pain frequency versus HUI3 pain, CHIP pain severity versus HUI3 pain, CHIP nonpain symptoms versus HUI3 dexterity, and CHIP psychosocial function versus HUI3 emotional and HUI3 cognitive.

## Results

### HUI3 HRQOL Data Analysis

HUI3 domain scores for CM-I patients are presented in Table 2. Measures of HRQOL related to vision, speech, cognition, and pain were all statistically lower in the CM-I population. The greatest outlier was the pain domain, in which the mean CM-I score was approximately 3 SDs below normal values, which is consistent with what is known about the pain features of CM-I. The exceptions to this are in measures of dexterity and ambulation, which were comparable. The mean multiattribute HUI3 composite score was  $0.70 \pm 0.32$  in CM-I patients (1.0 represents a “perfect” state of health). After ANOVA and post hoc testing of the CM-I patients only, HUI3 pain scores differed significantly between asymptomatic patients (1.00) and those with headaches (0.77) as well as between asymptomatic patients and those with paresthesias (0.82).

### CHIP Feasibility (Missing Items)

To assess the feasibility of administration, the percentage of administrations with missing values was calculated (Supplemental Table 1, accessible through link in endmatter). Within the physical domain, 16% of surveys had 1 or more missing items. Within the psychosocial domain, 15%

TABLE 2. Assessment of HRQOL in patients with CM-I via HUI3 (mean ± SD)\*

Item	Healthy†	All CM-I	p Value	Asymptomatic	Headaches Only	Paresthesias	p Value
Single-attribute HUI3							
Vision	0.99 ± 0.02	0.93 ± 0.20	0.04	1.00 ± 0.02 <sup>a</sup>	0.89 ± 0.24 <sup>a</sup>	0.99 ± 0.02 <sup>a</sup>	0.03
Hearing	1.00 ± 0.05	0.95 ± 0.23	0.88	1.00 ± 0.00	0.92 ± 0.23	0.88 ± 0.35	0.52
Speech	0.99 ± 0.07	0.91 ± 0.21	0.01	0.97 ± 0.06	0.85 ± 0.24	0.96 ± 0.12	0.18
Ambulation	1.00 ± 0.03	0.97 ± 0.14	0.31	1.00 ± 0.00	0.94 ± 0.17	1.00 ± 0.00	0.27
Dexterity	1.00 ± 0.04	0.99 ± 0.08	0.60	1.00 ± 0.00	1.00 ± 0.03	0.93 ± 0.19	0.49
Emotion	0.98 ± 0.05	0.92 ± 0.21	0.11	0.99 ± 0.04	0.89 ± 0.20	0.84 ± 0.34	0.36
Cognition	0.96 ± 0.09	0.88 ± 0.21	0.03	0.97 ± 0.05	0.85 ± 0.21	0.80 ± 0.30	0.13
Pain	0.97 ± 0.06	0.80 ± 0.22	<0.001	1.00 ± 0.07 <sup>a</sup>	0.77 ± 0.23 <sup>b</sup>	0.82 ± 0.16 <sup>b</sup>	0.001
Multiattribute HUI3							
Composite	—	—	—	0.94 ± 0.05 <sup>a</sup>	0.65 ± 0.30 <sup>b</sup>	0.64 ± 0.44 <sup>ab</sup>	<0.001

— = comparison was not made.

\* For CM-I 3-group comparison, means that do not share a superscript vary significantly on post hoc testing.

† From Pogany et al., 2006.

had 1 or more missing items. Overall, 25% of surveys had 1 or more missing items. The most common missing item was question number 13, “I have difficulty tying my shoelaces,” which was absent in 7% of cases. In reviewing these data and discussion with families, it appeared that this question was not answered because it was not applicable in cases where the child had not yet been taught to tie his/her shoes or the child did not wear shoes with shoelaces.

### CHIP Item Ranges and Normality

For most items, a full range of responses was demonstrated (Supplemental Table 1). When tested for normality, each of the 50 items was found to be not normally distributed. In general most item responses skewed toward 4 points each, indicating the best possible quality of life for that item.

### CHIP Scale Descriptives

Within the physical function domain, subscores ranged from 0.33 to 1.00 (Table 3). The mean score was 0.73 ± 0.16. The floor effect was 0% and the ceiling effect was 2%. Within psychosocial function, subscores ranged from 0.27 to 0.97 with a mean of 0.66 ± 0.20. The floor and ceiling effects were both 0%. The overall HRQOL scores ranged from 0.33 to 0.97, with a mean of 0.66 ± 0.18. The floor and ceiling effects were both 0%. The distribution

of scores was found to be not normal for all components. Each component of CHIP had roughly acceptable internal consistency, with Cronbach's alpha between 0.75 (pain severity) and 0.92 (psychosocial).

### CHIP Test-Retest Reliability

Ten patients (18%) met criteria for test-retest reliability analysis. The median time between administrations was 70 days (interquartile range 63–103 days). Secondary analyses revealed that these patients were a representative sample of the entire cohort. Distribution of age (13.1 ± 3.6 years), sex (50% male), symptoms (headache only, 80%; paresthesias, 20%), and radiology (syrinx, 20%) did not significantly vary from the entire cohort ( $p > 0.05$ ). Moreover, all baseline CHIP component scores were similar (all  $p > 0.37$ ). After this confirmation, test-retest analysis was performed (Table 3). For all components except pain frequency, Pearson's  $r$  exceeded 0.75 (range 0.81–0.94). Pearson's  $r$  for pain frequency was 0.62. For overall HRQOL, Pearson's  $r$  was 0.83.

### CHIP Construct Validity

CHIP construct validity was assessed in 3 ways: 1) comparison between CM-I and age-matched healthy controls, 2) comparison with symptomatology in CM-I, and 3) comparison against HUI3 in similar domains in CM-I.

TABLE 3. CHIP instrument characteristics

CHIP Component	Mean ± SD	Range	Floor Effect (%)	Ceiling Effect (%)	Feasibility (% complete)	Consistency (Cronbach's $\alpha$ )	Reliability (Pearson's $r$ )
Physical	0.66 ± 0.20	0.27–1.00	0	2	84	0.89	0.73
Pain frequency	0.59 ± 0.26	0.00–1.00	2	7	100	0.84	0.62
Pain severity	0.69 ± 0.20	0.19–1.00	0	7	95	0.75	0.83
Nonpain	0.82 ± 0.17	0.32–1.00	0	22	87	0.85	0.83
Psychosocial	0.68 ± 0.17	0.29–0.97	0	0	85	0.92	0.94
Overall HRQOL	0.66 ± 0.18	0.33–0.97	0	0	75	0.92	0.83

**TABLE 4. CHIP construct validity 1: patients with CM-I versus healthy controls (mean  $\pm$  SD)**

CHIP Component	Healthy	All CM-I	p Value	AUC
Physical	0.87 $\pm$ 0.11	0.66 $\pm$ 0.20	<0.001	0.809
Pain frequency	0.82 $\pm$ 0.15	0.59 $\pm$ 0.26	<0.001	0.776
Pain severity	0.87 $\pm$ 0.14	0.69 $\pm$ 0.20	<0.001	0.776
Nonpain	0.98 $\pm$ 0.04	0.82 $\pm$ 0.17	<0.001	0.806
Psychosocial	0.83 $\pm$ 0.11	0.68 $\pm$ 0.17	<0.001	0.754
Overall HRQOL	0.86 $\pm$ 0.10	0.66 $\pm$ 0.18	<0.001	0.820

### 1. Healthy Controls

Comparison of CHIP component scores between patients and controls is displayed in Table 4. The score for every component was lower in the patients versus controls (all  $p < 0.001$ ). Physical subscore was 0.87 in controls and 0.66 in CM-I (24% difference). Psychosocial subscore was 0.83 in controls and 0.68 in CM-I (18% difference). Overall HRQOL was 0.86 in controls and 0.66 in CM-I (23% difference). Physical and psychosocial scores achieved acceptable discriminative function, with AUC on ROC curve analysis of 0.809 and 0.754, respectively. Overall HRQOL discriminative function was 0.820.

### 2. Symptomatology

ANOVA comparing CHIP scores versus symptomatology is displayed in Table 5. The physical subscore demonstrated a significant difference across groups, with symptomatic patients having lower mean scores (headaches: 0.61, paresthesias: 0.55) compared with asymptomatic patients (0.90,  $p = 0.001$ ). There was a nonsignificant trend ( $p = 0.08$ ) toward lower psychosocial subscores in symptomatic patients as well. Overall HRQOL varied significantly (0.87 in asymptomatic patients vs 0.63 and 0.58 in headache and paresthesia groups, respectively,  $p < 0.001$ ).

### 3. CHIP versus HUI3

CHIP component scores were compared with relevant domains within the coadministered HUI3. The results of these analyses are presented in Table 6. Overall, CHIP scores correlated well with HUI3 scores. The physical subscore correlated well with the HUI3 pain ( $R^2 = 0.311$ ,  $p < 0.001$ ) metric. The psychosocial subscore correlated well with HUI3 cognitive ( $R^2 = 0.32$ ,  $p < 0.001$ ) and HUI3 emotional ( $R^2 = 0.16$ ,  $p = 0.003$ ) metrics. Overall HRQOL

correlated well with the HUI3 multiattribute composite score ( $R^2 = 0.440$ ,  $p < 0.001$ ).

## Discussion

A great challenge impeding efforts to improve the care of children diagnosed with CM-I is the lack of a standard outcome measure to evaluate pain, functionality, activities of daily living, and overall HRQOL. A better understanding of the relationship between HRQOL and clinical interventions will help inform future management of CM-I. Patient-reported outcome measurements have been applied in adults;<sup>12,30,31</sup> however, to date none have been evaluated in children. Recently, the CCOS has been shown to have a good interrater reliability and to correspond well with clinical gestalt assessment of outcome in children.<sup>44</sup> It is a rater-assessed scale assessing 4 dimensions: pain, non-pain symptoms, functionality, and complications.<sup>1</sup> While it is a practical clinical assessment tool, particularly in patients who undergo surgical treatment,<sup>16,28</sup> it does not use patient-reported outcomes and by itself does not establish a baseline of the severity of the patient's disease, such as measures like the Chiari Severity Index.<sup>13</sup>

To address this information gap, we developed the CHIP. It was determined that an effective HRQOL instrument in pediatric CM-I should meet the following criteria: 1) it should require patient involvement in answering the questions, 2) it should be age appropriate, 3) it should be thorough yet concise so that it can be completed in a short time period in the privacy of a patient room, and 4) it should help inform clinical decision making regarding treatment and management of these patients. Our experience thus far indicates that the CHIP has successfully met the first 3 criteria, and future studies are warranted to determine the clinical applications of this instrument.

In designing the CHIP, we have taken our cue from the efforts of Kulkarni et al. in their development of the HOQ.<sup>22,24</sup> The HOQ is a 51-point multidimensional HRQOL instrument that has been validated in children with hydrocephalus. While the authors of the HOQ initially reported intent for a discriminative index, later studies showed that it had capability of assessing outcomes as well (evaluative ability). In the 10 years since this instrument has been developed, several groups have adopted the HOQ to evaluate factors affecting HRQOL, as well as neurosurgical outcomes, in this population.<sup>21–23,25,26,32</sup> It is our hope that the CHIP may likewise be applied as an evaluative instrument in the pediatric CM-I population. A particular

**TABLE 5. CHIP construct validity 2: symptomatology in patients with CM-I (mean  $\pm$  SD)\***

CHIP Component	Asymptomatic	Headaches Only	Paresthesias	p Value
Physical	0.90 $\pm$ 0.08 <sup>a</sup>	0.61 $\pm$ 0.17 <sup>b</sup>	0.55 $\pm$ 0.22 <sup>b</sup>	0.001
Pain frequency	0.90 $\pm$ 0.11 <sup>a</sup>	0.53 $\pm$ 0.22 <sup>b</sup>	0.48 $\pm$ 0.28 <sup>b</sup>	<0.001
Pain severity	0.90 $\pm$ 0.15 <sup>a</sup>	0.65 $\pm$ 0.17 <sup>b</sup>	0.62 $\pm$ 0.23 <sup>ab</sup>	0.001
Nonpain	0.89 $\pm$ 0.15 <sup>a</sup>	0.82 $\pm$ 0.16 <sup>a</sup>	0.69 $\pm$ 0.21 <sup>a</sup>	0.045
Psychosocial	0.79 $\pm$ 0.15	0.66 $\pm$ 0.17	0.64 $\pm$ 0.21	0.08
Overall HRQOL	0.87 $\pm$ 0.08 <sup>a</sup>	0.63 $\pm$ 0.15 <sup>b</sup>	0.58 $\pm$ 0.21 <sup>b</sup>	<0.001

\* Means that do not share a superscript vary significantly on post hoc testing.

TABLE 6. CHIP construct validity 3: CHIP versus HUI3 (R2)\*

CHIP Component	HUI3				
	Overall	Pain	Dexterity	Cognitive	Emotional
Physical	0.380	0.311	0.082	—	—
Pain frequency	0.354	0.305	—	—	—
Pain severity	0.259	0.248	—	—	—
Nonpain	0.198	—	0.272	—	—
Psychosocial	0.344	—	—	0.324	0.155
Overall HRQOL	0.440	0.288	0.105	0.272	0.125

— = comparison was not made.

\* All correlations have p value < 0.05.

strength of the CHIP is that it may be administered at any time to establish a baseline HRQOL measurement before surgery, with subsequent systematic outcome assessments to follow, all from the patient's perspective.

### Application of the CHIP

There are 3 practical applications or abilities of HRQOL inventories: discriminative, evaluative, and predictive.<sup>14,19</sup> The discriminative ability of an index measures the capability of differentiating between patient groups and identifying meaningful differences in patient health status. An evaluative index is useful in the measurement of the magnitude of longitudinal change in an individual or group on the dimension of interest. A predictive index allows a patient to be assigned a risk category, which will inform necessity of intervention and/or type of intervention needed, and in some cases predict the response to an intervention.

A given HRQOL instrument may be beneficial in 1 or more of these 3 applications; however, instruments may excel in 1 area at the detriment to the others. The anticipated use of the instrument ultimately informs the design of the instrument, manifesting in various strengths and weaknesses of the inventory. The CHIP was designed to be useful in all 3 of these aspects, as discussed below. In this pilot study, we first assessed the discriminative ability of the CHIP in initial neurosurgical consultation of pediatric CM-I patients, as the latter 2 applications require additional longitudinal follow-up.

### Discriminative Ability

An important application of a discriminative index is in quantifying the disease burden across different communities of patients as well as differences between patients and healthy peers. Our initial experience has shown a high degree of feasibility, reliability, and validity in this population. Feasibility relates to the practical ability of participants to complete the survey. We found that 25% of returned surveys had missing items, the majority being nonpain related items that were not felt to be relevant by the patient. The scoring system used adjusts for this by scaling for completed responses only. We also found that all CHIP components were internally consistent, a measure of reliability of the test; the items in the psychosocial component might be somewhat redundant, as alpha (0.92) exceeded

the conventional 0.90 cutoff.<sup>35</sup> The physical and psychosocial components, as well as overall HRQOL score, were reliable, even over a median test-retest interval of 70 days.

Construct validity measures the extent to which a test measures what it claims to measure. We measured the differences between symptomatology of the CHIP and found significant differences between groups on the physical subscore. Additionally, we found that CHIP scores correlated with concurrent HUI3 scores within similar domains. Therefore, with regard to the discriminative abilities of the CHIP, we have demonstrated that it is a feasible, reliable, and valid assessment of HRQOL in pediatric CM-I.

### Evaluative Ability

This measure is useful for quantification of patient outcomes for comparative effectiveness research, as in clinical benefit or cost-effectiveness benchmarking in clinical trials. Within the area of pediatric CM-I, a HRQOL instrument with strong evaluative properties would be useful for comparing change in HRQOL with various treatment modalities. The present study has preliminarily assessed the discriminative abilities of the CHIP, but longitudinal assessment of the CHIP's evaluative properties, particularly its reliability, are ongoing in children who subsequently undergo surgical decompression with postoperative CHIP administration.

### Predictive Ability

Similarly, an index with strong predictive ability could be used in clinical decision making or patient counseling. Future studies of the CHIP will address the relationship between preoperative scores or individual item responses and outcomes following intervention.

### Limitations

The participants in this study were a nonrandom convenience sample of patients and parents willing to participate in the study. However, the population included is typical of the pediatric CM-I population with regard to age and radiographic features. Further, this single center study is being expanded to other centers to maximize the generalizability of the CHIP data. As external validation proceeds, we will also acquire more age-specific normative data that will help us to understand the usefulness of nonresponse questions.



## Conclusions

First, HUI3 data provide objective evidence that HRQOL in pediatric CM-I is markedly affected by pain. Second, we have presented some data to support the reliability and validity of the discriminatory abilities of this instrument, but further study is required and on-going. The CHIP is being implemented as a standard outcome tool at our institution. We are currently in the process of finalizing a federal grant, based on our accepted letter of intent, to evaluate surgical intervention, comparing risk and quality of life. Future studies of its evaluative and predictive properties in the neurosurgical management of CM-I are underway at ours and other centers across the United States in our collaborating network.

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## Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

## Author Contributions

Conception and design: Ladner, Wellons, Shannon. Acquisition of data: Ladner, Westrick. Analysis and interpretation of data: all authors. Drafting the article: Ladner. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Ladner. Statistical analysis: Ladner, Westrick, Shannon. Administrative/technical/material support: Wellons, Shannon. Study supervision: Wellons, Shannon.

## Supplemental Information

### Online-Only Content

Supplemental material is available with the online version of the article.

*Chiari Health Index for Pediatrics and Supplemental Table 1.* <http://thejns.org/doi/suppl/10.3171/2015.5.PEDS1513>.

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