Medical, Social, and Economic Factors Associated with Health-Related Quality of Life in Canadian Children with Hydrocephalus

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Objective To study the factors associated with health-related quality of life (HRQL) in Canadian children with hydrocephalus, using a comprehensive model of determinants of child health, including socioeconomic factors.

Study design A cross-sectional study was performed between November 2005 and November 2006 at 3 Canadian pediatric hospitals. Parents of children with hydrocephalus age 5 to 18 years completed the Hydrocephalus Outcome Questionnaire (HOQ) and the Health Utilities Index Mark 3 (HUI-3).

Results A consecutive sample of 340 subjects participated from a total of 366 eligible children (mean age, 11.6 ± 3.6 years; mean time from the diagnosis of hydrocephalus, 10.0 ± 4.6 years). Adjusted multivariate linear regression models demonstrated that the most important determinants of poorer HRQL included lower family income, lower parental education, worse family functioning, seizures, myelomeningocele, and prolonged treatment for cerebrospinal fluid shunt obstruction.

Conclusions Despite a national universal health care system, socioeconomic disparities remain important as determinants of HRQL. Given the absence of a parallel private health care system in Canada, this suggests that the impact of socioeconomic factors is related to issues other than access to care. *(J Pediatr 2008;xx:xxx)*

Health care in Canada is predominately publicly funded and governed under the Canada Health Act, which states as its primary objective "to protect, promote and restore the physical and mental well-being of residents of Canada and to facilitate reasonable access to health services without financial or other barriers."[1] No parallel private insurance is allowed for publicly insured services. Children with chronic illness would appear to benefit most from such a system because of their increased need for costly medical care. An example of this is pediatric hydrocephalus, a common serious chronic condition.[2] These children are treated acutely with a cerebrospinal fluid (CSF) shunt, typically a ventriculoperitoneal shunt. For the thousands of children born annually with such conditions as prematurity, myelomeningocele (spina bifida), and other congenital anomalies, the development of hydrocephalus and its management (including recurring neurosurgery for CSF shunt malfunction) present major challenges later in life, requiring multidisciplinary care from pediatricians, neurosurgeons, neurologists, rehabilitative therapists, and others.

The goals of the present study were to quantify long-term health-related quality of life (HRQL) outcomes in children with hydrocephalus many years after their initial treatment, using a reliable validated outcome measure, and also to identify factors associated with outcome in a large, diverse sample of children from centers across Canada, all of whom were treated under a national universal health care program. We used a conceptual framework recently articulated by Stevens,[3] borrowing earlier concepts from others,[4] that defines 5 domains of child health influences: biology, family structure, socioeconomic status, family/community environment, and health care. We were interested in investigating the extent to which socioeconomic disparities are important in a publicly funded health care system.

CSF	Cerebrospinal fluid	HOQ	Hydrocephalus Outcome Questionnaire
GF-FAD	General Functioning Scale of the McMaster Family Assessment Device	HUI-3	Health Utilities Index Mark 3
		HRQL	Health-related quality of life

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METHODS

A consecutive sample of children age 5 to 18 years with a diagnosis of hydrocephalus treated at least 6 months earlier was recruited from the neurosurgery outpatient clinics at 3 tertiary pediatric hospitals across Canada (Sick Kids, Toronto; BC Children's Hospital, Vancouver; and IWK Health Centre, Halifax) between November 2005 and November 2006. This sample was typical of the childhood hydrocephalus population, because virtually all children with hydrocephalus in Canada are treated and seen annually by pediatric neurosurgeons at tertiary care pediatric hospitals. The study protocol was approved by the Institutional Review Boards of The Hospital for Sick Children, University of British Columbia, and IWK Health Centre, and written consent was obtained from all participating families.

Assessment of Medical, Social, and Economic Factors

The medical and imaging history was reviewed for each subject to identify several potential medical factors considered potentially relevant to outcome: current age, age at first treatment for hydrocephalus,[5,6] preterm birth (< 38 weeks gestation),[7] underlying diagnosis of myelomeningocele,[8] total number of days spent in the hospital for initial treatment of hydrocephalus, average annual length of stay for CSF shunt infection,[9] total number of hospital admissions,[8-10] average annual length of stay for CSF shunt obstruction,[8-10] current ventricular size[11] (measured from brain magnetic resonance imaging or computed tomography using the frontal-occipital horn ratio, a previously validated linear measure of ventricular size),[12,13] and presence of seizures (having had at least 1 seizure episode in the previous 12 months).[8,14-17]

Data on several social and economic factors also were collected from the primary caregiver, including whether at least 1 parent had earned a university degree, total annual household income > $100 000 (which approximates the top 20% of family incomes in Canada[18]), family structure (2-parent vs other), whether at least 1 parent was employed full time, and whether the child's mother was born in Canada. Family functioning was measured using the General Functioning Scale of the McMaster Family Assessment Device (GF-FAD).[19,20] The GF-FAD, which has proven to be reliable and valid, is based on a model of family functioning that incorporates multiple dimensions, including problem solving, communication, roles, affective responses, affective involvement, and behavior control. GF-FAD scores range from 1 to 4, with higher scores indicating greater pathology in family functioning. Geographic access to tertiary care was determined by calculating the driving distance from the family residence to the pediatric hospital.[21] The large geographic areas served by each hospital can make the distance from tertiary care a potential barrier for some patients, especially by causing delays in urgent surgical intervention, such as an acute CSF shunt obstruction. As a measure of neighborhood affluence, median family income for the geographic region (based on postal code) in which the child lived was obtained from Canadian census data.

Together, these variables addressed all 5 domains of child health influence proposed by Stevens:[3] biology (age, age at first treatment, myelomeningocele, preterm birth, seizures, ventricular size), family structure (2-parent family), socioeconomic status (maternal nativity, family income, parental education, parental employment), family/community environment (family functioning using the GF-FAD, distance from pediatric hospital, median neighborhood family income), and health care (total number of days spent in hospital for initial treatment of hydrocephalus, average annual length of stay for CSF shunt infection, total number of hospital admissions, average annual length of stay for CSF shunt obstruction).

Assessment of HRQL

The child's primary caregiver completed the Hydrocephalus Outcome Questionnaire (HOQ), a 51-item questionnaire with proven reliability and validity in measuring health outcomes in children with hydrocephalus.[16,22] We have previously reported good correlations between the HOQ and several independent measures of health,[22] including the Wide-Range Achievement Reading Test,[23] Strengths and Difficulties Questionnaires,[24] and Functional Independence Measure for Children.[25] The HOQ provides scores of Overall Health, Physical Health, Cognitive Health, and Social-Emotional Health, all of which range from 0 (worse outcome) to 1.0 (better outcome). Previous work has suggested that to be clinically meaningful, a difference in HOQ scores needs to be approximately on the order of 0.10 or greater, based on how parents perceive differences in health status.[26] Because the HOQ is a disease-specific instrument, there are no healthy population data for the HOQ. Thus, to provide a general population comparison, caregivers also completed the Health Utilities Index Mark 3 (HUI-3). This provided utility scores that could be compared with established population norms and other patient populations, to help impart more clinically relevant meaning to a given health state.[26-29] Utility scores from the HUI-3 can be < 0, indicating states described as "worse than dead."[29]

Statistical Analysis

A multivariate least squares linear regression model was built using the HOQ Overall Health score as the dependent variable. A backward selection process was used, retaining independent variables with $P < .10$ in the multivariate model. An a priori decision was made to retain the following variables in all multivariate models based on clinical suspicion that they could be potentially important confounders: age, age at initial treatment of hydrocephalus, and diagnosis of myelomeningocele. All analyses were adjusted for study center by including study center as a variable in all models. The variance inflation factor of all independent variables was < 10, suggesting that multicollinearity was not a concern.[30] A P value < .05 in the multivariate model was taken to suggest a significant association between the independent variable and outcome. Separate multivariate models were built using each of the HOQ

Table I. Patient characteristics

Category	Characteristic	Value
Biological	Age at assessment, years, mean/median (SD)	11.6/12.1, (3.6)
	Age at first surgery, months, mean/median (SD)	19.5/2.1, (37.3)
	Myelomeningocele, n (%)	112 (32.9%)
	Premature birth, n (%)	90 (29.5)
	Seizures, n (%)	52 (15.4)
	Current ventricular size, frontal-occipital horn ratio, mean/median (SD)	0.43/0.43 (0.07)
Family structure	Two-parent family, n (%)	272 (80.5)
Socioeconomic status	Reported annual household income >$100,000 Canadian dollars, n (%)	52 (16.9)
	Mother born in Canada, n (%)	242 (71.6)
	At least 1 parent completed university degree, n (%)	51 (15.3)
	At least one parent employed full-time, n (%)	75 (83.3)
Family/community environment	Family functioning, McMaster Family Assessment Device score, mean/median (SD)	1.55/1.50 (0.45)
	Median neighborhood family income, Canadian dollars, mean/median (SD)	61 420/60 000 (17,153)
	Distance of family residence from pediatric hospital, km, mean/median (SD)	134/58 (195)
Health care	Total number of days spent in hospital for initial treatment of hydrocephalus, mean/median (SD)	17.9/10.0 (21.6)
	Total number of hospital admissions, mean/median (SD)	2.1/1.0 (3.1)
	Average annual length of hospital stay for treatment of CSF shunt infection, days/year, mean/median (SD)	0.3/0.0 (1.4)
	Average annual length of hospital stay for treatment of CSF shunt obstruction, days/year, mean/median (SD)	0.8/0.2 (1.8)

*The frontal-occipital horn ratio is approximately 0.37 for normally sized ventricles and generally > 0.55 for severe ventriculomegaly.[12,13]

subscores (Physical Health, Cognitive Health, and Social-Emotional Health) as the dependent variable. Plots of residuals were examined for each model to check assumptions of linearity and normality. All analyses were performed using SPSS Advanced Statistics 13.0 (SPSS Inc, Chicago, Illinois).

RESULTS

A total of 366 patients were approached to participate in this study. Of these, 26 (7.1%) did not complete the required questionnaires, usually due to time constraints. The 340 patients who completed the questionnaires (244 from Toronto, 58 from Vancouver, and 38 from Halifax) formed the basis of our analysis. Based on estimated annual incidence data, this cohort represents about 5% of the Canadian hydrocephalus population between age 5 and 18 years.[2]

Only 5 predictor variables had > 3% missing data: premature birth (10.3% missing), age at first treatment for hydrocephalus (9.3% missing), average annual length of hospital stay for treatment of CSF shunt infection (8.8% missing) or CSF shunt obstruction (10.6% missing), and reported total annual household income (9.4% missing). The missing data for the first 4 variables was due to the unavailability of old medical records. There was no statistically significant difference in patient age, family functioning score, or any of the outcome variables between those with complete versus missing data for all 5 variables (all $P > .05$; independent sample t-test). Moreover, there was no difference in any of the other social and economic variables (parental education, parental employment status, and maternal nativity) between those who reported their income and those who did not (all $P > .05$; χ^2 test).

Table II. Outcome assessment

Outcome	Value
HOQ Overall Health Score, mean/median (SD)	0.65/0.67 (0.20)
HOQ Physical Health Score, mean/median (SD)	0.66/0.68 (0.25)
HOQ Cognitive Health Score, mean/median (SD)	0.55/0.56 (0.28)
HOQ Social-Emotional Health Score, mean/median (SD)	0.71/0.75 (0.19)
HUI-3 Utility Score, mean/median (SD)	0.58/0.63 (0.32)

Patient characteristics and outcome assessments are given in Tables I and II and the Figure. Aside from myelomeningocele (112; 32.9%), other common underlying etiologies included aqueduct stenosis (33; 9.7%), intraventricular hemorrhage of prematurity (32; 9.4%), posterior fossa cyst (17; 5.0%), and postinfection (15; 4.4%). In the HUI-3 domains, impairments in the following areas were reported: cognition in 61.1%, ambulation in 51.1%, pain in 35.8%, vision in 33.2%, speech in 29.2%, emotions in 25.2%, dexterity in 22.3%, and hearing in 5.3%. Seventeen subjects (5.0%) had HUI-3 utility scores < 0.

Determinants of HRQL

The multivariate analysis for determinants of HOQ Overall Health score yielded the following significant associations with *worse* outcome (adjusted $R^2 = 0.26$): seizures, worse family functioning, myelomeningocele, total household income < $100 000, and no parent with a university degree (Table III). The results of the multivariate analyses for the HOQ subscores are given in Table III. The adjusted R^2

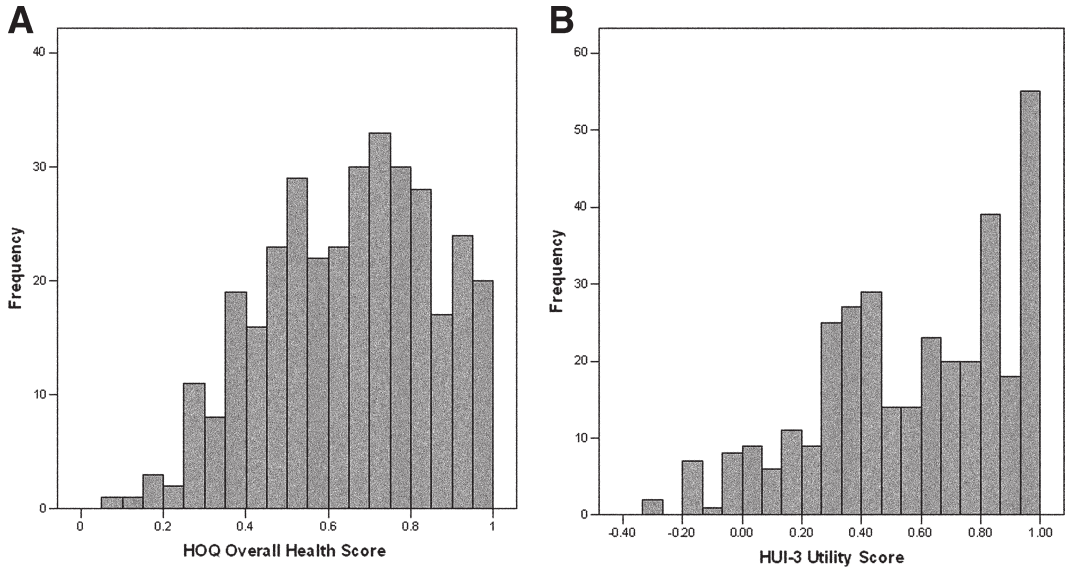

Figure. Histograms showing the distribution of **A,** HOQ scores and **B,** HUI-3 scores.

values were 0.30 for Physical Health, 0.18 for Cognitive Health, and 0.18 for Social-Emotional Health.

DISCUSSION

In our study of 340 children from centers across Canada, we found that the long-term outcome in pediatric hydrocephalus was highly variable, with wide-ranging determinants of outcome. These include socioeconomic status (eg, family income, parental education), family environment (eg, family functioning), biology (eg, seizures, myelomeningocele, age), and health care (eg, length of stay for treatment of CSF shunt obstruction). The effect of most, but not all, of these variables appeared to be clinically meaningful as well, with effects of >0.1 on the HOQ score (Table III).[26] Our findings provide unique insight into the relative importance of these factors, particularly in the setting of universal health care in Canada.

In countries without universal health care, such as the United States, the effect of socioeconomic status on child health outcomes has been well demonstrated; for example, lower income has been associated with greater unmet health care needs,[31,32] worse overall child health outcomes,[33] and poorer social and cognitive outcomes in young children.[34] In Canada, the goal of publicly funded health care is to eliminate the effects of socioeconomic disparities; nonetheless, individual socioeconomic advantage has been demonstrated to be of some benefit in accessing services for adult cardiac patients,[35] improving birth outcomes,[36] receiving inhaled steroid therapy for childhood asthma,[37] and preventing toddler health problems.[38] At a community level, lower neighborhood socioeconomic status has been associated with a higher prevalence of childhood obesity[39] and greater childhood injury-related mortality.[40]

We have found that despite universal health care coverage, there is a clear and important health outcome advantage for children from high-income families (in the top 20% of Canadian families). Although the Canadian health care system removes financial barriers to accessing primary medical care and any necessary tertiary medical care (including surgery, hospitalizations, and diagnostic imaging), some medical costs are not covered, including prescription medications, dental care, some rehabilitation therapy services, and some travel costs to tertiary centers, which can be substantial for those living in remote communities. Lower family income may present barriers to this type of care.

We also found that higher parental education is an important predictor of better child overall outcome and cognitive outcome. This concurs with previous reports demonstrating associations between higher parental education and improved access to care,[41] improved outcome in children with brain tumors[42] and sickle cell disease,[43] as well as in the National Health Interview Survey.[44]

The importance of the family unit and parent–child interactions to overall child health has been well documented.[4] In our study, we found a strong and clear association between worse family functioning (ie, higher GF-FAD score) and worse child HRQL in all dimensions. Although it may be that inherently worse family functioning adversely affects the health outcome of children with hydrocephalus, it is also possible that family functioning deteriorates secondarily in families with children in poorer health. We found a strong association between the presence of epilepsy and poor outcome, confirming previous reports.[8,14-17] Myelomeningocele was associated with a worse outcome; not surprisingly, this effect was most pronounced in the physical domain. Older age was found to a significant variable related to better physical outcome only; however, given the small magnitude of this effect, this may represent a spurious finding. Our findings suggest that those children who required prolonged and repeated treatment of CSF shunt obstructions fared worse. This

Table III. Association of patient and family characteristics with outcome

Category	Variable	Unstandardized regression coefficient (95% confidence interval)*			
		HOQ Overall Health Score	HOQ Physical Health Score	HOQ Cognitive Health Score	HOQ Social-Emotional Health Score‡
Socioeconomic status	Household income >$100 000/year	0.08 (0.03 to 0.14) $P = .006$	0.10 (0.04 to 0.17) $P = .003$	0.11 (0.03 to 0.19) $P = .01$	0.07 (0.02 to 0.13) $P = .01$
	At least 1 parent completed university degree	0.06 (0 to 0.12) $P = .047$	NA	0.14 (0.05 to 0.22) $P = .002$	NA
Family/community	Family functioning (McMaster GF-FAD score)†	−0.14 (−0.18 to −0.09) $P < .001$	−0.08 (−0.13 to −0.03) $P = .003$	−0.14 (−0.21 to −0.08) $P < .001$	−0.14 (−0.18 to −0.09) $P < .001$
Biological	Seizures	−0.14 (−0.20 to −0.08) $P < 0.001$	−0.21 (−0.27 to −0.14) $P < .001$	−0.13 (−0.21 to −0.04) $P = .004$	−0.07 (−0.13 to −0.01) $P = .03$
	Myelomeningocele	−0.07 (−0.12 to −0.02) $P = .009$	−0.15 (−0.21 to −0.09) $P < .001$	NA	NA
	Age at assessment, years	NA	0.01 (0 to 0.02) $P = .02$	NA	NA
Health care	Average annual length of hospital stay for treatment of CSF shunt obstruction, days/year	NA	−0.02 (−0.03 to 0) $P = .03$	NA	NA

NA, not applicable because the variable either was not included in the multivariable model or did not reach statistical significance ($P < .05$).

*Regression coefficients from multivariable linear regression corrected for all other variables in the model.

†GF-FAD scores range from 1 to 4; a higher score implies worse family functioning.

‡There was also a significant association between medical center and HOQ Social-Emotional Health Score (see the text).

could be related to the pathophysiologic effect of abnormally high intracranial pressure on the developing brain or to the effect of a prolonged time away from school and other normal activities.

Although the outcome in children with hydrocephalus is highly variable, the cognitive scores were notably lower than either the physical or social-emotional scores, and a majority of the subjects (61.1%) reported impaired cognitive ability on the HUI-3. Previous neuropsychological studies in children with hydrocephalus have revealed difficulties with reading comprehension, oral comprehension, narrative discourse, and math skills, perhaps related to slow information processing skills and visual-spatial deficits.[45-50] Perhaps of most concern, however, is the substantial overall HRQL burden carried by this population; their mean utility score was only 0.58 ± 0.32 (compared with, eg, the 0.89 reported for a sample of control Canadian children,[29] 0.87 for adolescents born at extremely low birth weight,[51] and 0.58 for adults with Alzheimer's disease[52]), and 5.0% had scores indicating a state "worse than dead."

Our study has some limitations. The assessment of some independent variables was retrospective. Although this assessment was done by trained research personnel reviewing the subjects' complete medical records, the potential for inaccuracies and missing data remained. We found no systematic differences in any relevant variables between those patients with complete data and those with missing data, suggesting no inherent bias in our data sample. Although the sample is large and comes from multiple centers across geographically diverse regions of Canada, the ability to generalize our findings to other countries, especially those without universal health care, is limited. As well, by sampling those subjects who attended their clinic visits, we might have missed some who were doing either very well or very poorly and were less motivated to attend the clinic. Nonetheless, the characteristics of our sample in terms of age, underlying etiology, and disease severity appear to be typical and representative of children with hydrocephalus. The utilities scores calculated from the HUI-3 represent the preferences of the general population and thus may not adequately represent the perspective of the patients or affected families themselves. Moreover, all health outcomes were measured using parental proxy respondents, which possibly could underestimate a child's perspective of his or her own health.[53] Despite the fact that we considered several seemingly important predictor variables, the amount of variance explained by any of our multivariate models was relatively low (none exceeding an adjusted R^2 of 0.30). This suggests that there remains a large amount of variance in health outcomes that we cannot explain, in keeping with the findings of similar outcome studies in other disease conditions.[54-56] It is hoped that future research will improve our understanding of the complex interactions that determine a child's ultimate outcome.

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