Childs Nerv Syst
DOI 10.1007/s00381-010-1162-6

ORIGINAL PAPER

Does treatment with endoscopic third ventriculostomy result in less concern among parents of children with hydrocephalus?

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Received: 13 April 2010 / Accepted: 16 April 2010

Abstract
Purpose A possible benefit of endoscopic third ventriculostomy (ETV) is that families might harbor less concern and anxiety compared to shunt. This has not yet been demonstrated, however. Our goal was to compare parental concern in a large sample of children with hydrocephalus treated with ETV or shunt, using our previously developed measure of parental concern, the Hydrocephalus Concerns Questionnaire for Parents (HCQ-P).
Methods The parents of children 5–18 years old with previously treated hydrocephalus at three Canadian pediatric neurosurgery centers completed the HCQ-P. HCQ-P scores were compared between those who were initially treated with ETV and those initially treated with shunt. A multivariable linear regression analysis was used to adjust for center, current age, age at initial hydrocephalus surgery, seizures, etiology, hydrocephalus complications, and quality of life.
Results Six hundred three families participated (58 ETV [9.6%], 545 shunt [90.4%]). In unadjusted comparison, ETV parents had lower overall concern (HCQ-P=0.41 versus 0.51, p=0.02). After adjustment for multiple patient factors, ETV parents still had lower concern (p=0.03) but the only questions for which there was a still a statistically significant difference were those related to concerns about shunt/ETV complications.
Conclusions Parents of children who have had ETV experience less concern than those who have had shunt and this is due almost exclusively to less concern about hydrocephalus treatment complications. While this could be interpreted as a beneficial aspect of ETV treatment, it remains important for neurosurgeons to ensure that parents are not being overly complacent about the possibility of ETV failure requiring urgent treatment.

Keywords Hydrocephalus · Pediatric · Health outcome · Parental concern · Quality of life

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Introduction

The purported benefits of endoscopic third ventriculostomy (ETV) over cerebrospinal fluid (CSF) shunt in the treatment of childhood hydrocephalus cover many potential spheres. One important one is the possibility that parents and patients might experience less anxiety and concern about their child's condition and future after treatment with ETV compared to CSF shunt. This issue has not, to our knowledge, previously been studied. Our objective with this study was to examine specifically the relationship between parental concern and treatment with either ETV or shunt, using our previously developed Hydrocephalus Concerns Questionnaire for Parents (HCQ-P) [6, 7] as the primary measure. We hypothesized that, even after correcting for other patient factors, parents of children treated initially with ETV would experience less concern about their children than those treated with shunt.

Published online: 29 April 2010

Clinical materials and methods

Between July 2005 and August 2008, all patients with hydrocephalus attending a regularly scheduled neurosurgery out-patient clinic at Sick Kids, Toronto were eligible to participate in this study. During portions of this time, patients were also recruited at B.C. Children's Hospital and IWK Health Centre. All children with a history of surgically treated hydrocephalus, which was initially treated at least 6 months earlier, and were between the ages of 5 and 18 years were eligible.

The medical charts were reviewed by the research team to identify several factors related to the child's surgical history. We concentrated on factors that we have previously shown to be associated with parental concern [7]. Etiology of hydrocephalus was initially classified as one of the following: post-infectious (meningitis or abscess), post-intraventricular hemorrhage secondary to prematurity, myelomeningocele, aqueductal stenosis, spontaneous intracranial hemorrhage (e.g., arteriovenous malformation, aneurysm), posterior fossa tumor, supratentorial tumor, midbrain tumor or other midbrain lesion (e.g., pineal cyst), post-head injury, encephalocele, posterior fossa cyst, other intracranial cyst (e.g., arachnoid cyst, porencephalic), communicating congenital hydrocephalus, other congenital (e.g., schizencephaly, holoprosencephaly), craniosynostosis, or other. For the purposes of statistical analyses, all etiological categories with fewer than 20 patients were added to the "other" category. In order to capture both the frequency and the severity of surgical complications, the *total number of days* the child was admitted to hospital for the initial treatment of hydrocephalus and any subsequent hydrocephalus-related complications over the child's lifetime was recorded. This was felt to more accurately reflect the complexity and severity of the child's initial presentation and future complications.

Quality of life (QOL) was measured using the Hydrocephalus Outcome Questionnaire (HOQ) [4, 5]. Parents were asked to complete the HOQ by a trained research assistant. The HOQ is a reliable and previously validated measure of QOL with scores ranging from 0 (worst QOL) to 1.0 (best QOL). The HOQ provides scores of Overall Health, Physical Health, Cognitive Health, and Social-Emotional Health. The responses from the child's mother or father (if mother was not available) were used for all data analyses. We have previously shown good agreement between mother and father HOQ responses [4].

We collected socio-economic data from participating families during a portion of our study period, so this data was only available for a subset of the larger sample. The following data was collected by parental self-report: household income, number of family members, parental education (did either parent graduate from university?), parental employment status (was either parent fully employed?).

Outcome measure Parents completed the HCQ-P, which is a measure of concern of parents of children with hydrocephalus. The HCQ-P is a nine-item questionnaire that asks about a parent's concern about their child's future in various aspects of life. All begin with the stem, "I am concerned..." and conclude with the nine items shown in the first column of Table 2. Three questions asked about shunt complications specifically (Q4, Q8, and Q9). These were re-worded for parents of children with ETV to make these questions relevant to them, e.g., "...third ventriculostomy will become blocked" instead of "...shunt will become blocked". We have previously demonstrated the reliability of the HCQ-P and shown evidence of construct validity [6, 7]. The HCQ-P provides a score that ranges from 0 (least parental concern) to 1 (most parental concern).

Data analysis We used independent sample *t* tests to compare the responses to each of the nine questions in the HCQ-P and total HCQ-P score between patients initially treated with shunt versus those initially treated with ETV (even if the ETV failed and the child was eventually shunted).

To account for differences in patient characteristics and complications, we performed a multivariable linear regression analysis in which maternal HCQ-P score was the dependent variable. In addition to initial hydrocephalus treatment (ETV vs. shunt), we adjusted for the following possible confounders: center of treatment (Toronto, Vancouver, Halifax), current age, age at initial hydrocephalus surgery, frequency of seizures, etiology of hydrocephalus, total days spent in hospital for initial treatment of hydrocephalus, total number of days spent in hospital for treatment of subsequent hydrocephalus complications, and measures of quality of life (HOQ Physical Score, Cognitive Score, and Social-Emotional Score). Because of the skewed distribution of days in hospital, these variables were log-transformed for analysis. Variance inflation factors for all independent variables were less than 3, suggesting that multicollinearity was not a concern. We performed separate multivariable linear regression models for responses to each of the nine HCQ-P questions as the dependent variable. We repeated the analysis using the presence of a functioning ETV at assessment versus functioning shunt at assessment to determine whether the results would differ substantially. All analyses were repeated using paternal responses as the dependent variable. A further set of analyses were carried to include adjustment for family socio-economic indicators (household income,

parental education, parental employment status, number of family members), recognizing that this data was only available for a subset of patients.

All analyses were performed using SPSS Advanced Statistics 17.0 (SPSS Inc., Chicago, IL, USA). A p value of <0.05 was taken to suggest an association of interest between ETV/shunt and parental concern.

Participants in this study were compensated for their time with a small monetary gift. This protocol was approved at the coordinating center by the Sick Kids Research Ethics Board (number 1000005376) and at each participating center.

Results

Data from 603 patients was available for analysis (478 from Toronto, 89 from Vancouver, and 36 from Halifax). The overall participation rate in this study was over 80% of those approached. Fifty-eight (9.6%) patients had ETV as their primary procedure, while 545 (90.4%) had shunt. The descriptive data of this sample are shown in Table 1. These showed the expected differences between the ETV and shunt population.

A raw comparison of maternal responses is shown in Table 2. There was much less overall concern among mothers of ETV patients than mothers of shunt patients (HCQ-P Score 0.41 vs. 0.51, p=0.02). This was consistent across most of the nine questions of the HCQ-P. The only questions for which there was no statistical difference were questions related to concerns about the child's future education (Q5), ability to maintain friendships (Q6), and ability to participate in sports (Q7).

To adjust for the possible impact of other patient factors on parental concern, we used a linear regression model in which the effect of ETV versus shunt treatment was adjusted for all factors listed in Table 1 and center of treatment (Toronto, Vancouver, or Halifax). The results, presented as unstandardized regression coefficients, are shown in Table 3. These regression coefficients represent

Table 1 Patient characteristics

Variable	ETV as first treatment (N=58)	Shunt as first treatment (N=545)	p value
Current age (years)	10.4 (3.5)	11.9 (3.8)	0.004
Age at first surgery (months)	66.9 (57.1)	17.0 (36.6)	<0.001
Etiology of hydrocephalus			
Myelomeningocele	2 (3%)	174 (32%)	<0.001
Aqueduct stenosis	18 (31%)	50 (9%)	
Post-infectious	1 (2%)	24 (4%)	
IVH of prematurity	1 (2%)	73 (13%)	
Congenital communicating	0	28 (5%)	
Posterior fossa cyst	2 (3%)	30 (6%)	
Midbrain lesion	10 (17%)	0	
Tumor (non-midbrain)	14 (24%)	54 (10%)	
Other	10 (17%)	112 (21%)	
Frequency of seizures			
Never	48 (83%)	452 (83%)	0.93
<1/year	5 (9%)	38 (7%)	
1–12/year	2 (3%)	21 (4%)	
2–4/month	0	10 (2%)	
2–7/week	1 (2%)	8 (1%)	
≥2/day	2 (3%)	16 (3%)	
Number of days spent in hospital for initial admission	17.4 (48.3)	17.6 (19.2)	0.97
Number of days spent in hospital for hydrocephalus complications	5.4 (14.4)	16.5 (29.9)	0.005
Presence of functioning shunt at time of assessment	25 (43%)	536 (98%)	<0.001
HOQ Physical Health Score	0.79 (0.19)	0.69 (0.24)	0.002
HOQ Cognitive Health Score	0.58 (0.30)	0.56 (0.28)	0.60
HOQ Social-Emotional Health Score	0.73 (0.21)	0.72 (0.18)	0.70
HOQ Overall Score	0.70 (0.22)	0.66 (0.20)	0.12

Values presented are either mean (standard deviation) or absolute number (percentage)

CSF cerebrospinal fluid, *ETV* endoscopic third ventriculostomy, *FOR* frontal-occipital horn ratio, *IVH* intraventricular hemorrhage

Table 2 Unadjusted comparison of HCQ-P maternal responses

Outcome variable	ETV as first treatment	Shunt as first treatment	*p* value
HCQ-P total score	0.41 (0.31)	0.51 (0.29)	0.02
Q1. About my child's ability to take care of a family in the future	2.4 (1.5)	2.9 (1.6)	0.04
Q2. About my child's ability to find a career	2.6 (1.6)	3.1 (1.7)	0.04
Q3. About my child's ability to live alone in the future	2.4 (1.6)	2.9 (1.7)	0.03
Q4. About the need for future (hydrocephalus or shunt) surgery	2.9 (1.7)	3.5 (1.4)	0.005
Q5. About my child's future education	3.1 (1.7)	3.3 (1.6)	0.42
Q6. About my child's ability to maintain friendships	2.4 (1.7)	2.5 (1.6)	0.70
Q7. About my child's ability to participate in sports in the future	2.6 (1.5)	2.7 (1.5)	0.76
Q8. That my child's (third ventriculostomy or shunt) will become blocked	2.4 (1.6)	3.4 (1.5)	<0.001
Q9. That my child will get a (brain fluid (CSF) or shunt) infection	2.3 (1.6)	3.3 (1.5)	<0.001

Values presented are mean (standard deviation). In all cases, higher values represent more concern. The HCQ-P total score is on a scale from 0 (least concern) to 1.0 (most concern) and the responses to Q1 to Q9 are on a scale from 1 (least concern) to 5 (most concern).

HCQ-P Hydrocephalus Concerns Questionnaire for Parents

the average difference between maternal responses for ETV patients and shunt patients, once all other factors are corrected for. Almost all values were negative, again indicating less concern among the mothers of ETV patients. With these factors taken into account, however, the only questions for which there was a still a statistically significant difference were those related to concerns about shunt/ETV complications (Q4, Q8, and Q9). Overall concern also remained less in the ETV group than the shunt group.

At the time of assessment, only 42 patients had functioning ETV (33 initially treated with ETV and nine initially treated with shunt). We repeated the analyses to assess whether the presence of a functioning ETV at assessment would alter the results, but, in fact, the results were very similar (data not shown). When paternal responses were used instead of maternal responses, the results were also similar (data not shown).

Family socio-economic data was available for 333 patients (40 ETV, 293 shunt). There was no significant difference in the socio-economic indicators between the family of ETV patients and shunt patients: household income ($p=0.95$, t test), parental education ($p=0.41$, chi-square), parental employment status ($p=0.59$, chi-square), number of family members ($p=0.54$, t test). Within this subset of 333 patients, a multivariable linear

Table 3 HCQ-P responses adjusted comparison

Outcome variable	Unstandardized regression coefficient for ETV as first treatment (95% confidence interval)	*p* value
HCQ-P total score	−0.08 (−0.16 to −0.01)	0.03
Q1. About my child's ability to take care of a family in the future	−0.29 (−0.70 to 0.13)	0.17
Q2. About my child's ability to find a career	−0.36 (−0.77 to 0.04)	0.08
Q3. About my child's ability to live alone in the future	−0.28 (−0.70 to 0.13)	0.18
Q4. About the need for future (hydrocephalus or shunt) surgery	−0.63 (−1.1 to −0.14)	0.01
Q5. About my child's future education	−0.09 (−0.50 to 0.31)	0.64
Q6. About my child's ability to maintain friendships	0.02 (−0.40 to 0.36)	0.91
Q7. About my child's ability to participate in sports in the future	−0.25 (−0.69 to 0.19)	0.26
Q8. That my child's (third ventriculostomy or shunt) will become blocked	−0.91 (−1.42 to −0.39)	0.001
Q9. That my child will get a (brain fluid (CSF) or shunt) infection	−0.77 (−1.30 to −0.23)	0.005

Values presented are unstandardized regression coefficients (95% confidence intervals), as derived from a multivariable linear regression model with adjusted for several patient variables (see text). Negative values indicate that parents of children who had ETV as their initial procedure had less concern than the parents of initially shunted children

HCQ-P Hydrocephalus Concerns Questionnaire for Parents

regression model was performed that adjusted for family socio-economic indicators in addition to the other confounders. The results were similar to the main analysis, with less concern among mothers of ETV patients than mothers of shunt patients (data not shown).

Discussion

We have shown that, in a large sample of children with hydrocephalus, overall parental concern appears to be less in ETV patients than shunt patients. Once numerous patient-related factors are corrected for, it appears that the areas in which ETV parents feel less concern involve hydrocephalus complications and the need for future surgery.

The main strength of our study is its large sample size. This allowed for more than adequate statistical power with which to perform an adjusted regression analysis. We used a measure of parental concern with proven reliability and evidence of construct validity that was developed specifically for, and by, parents of children with hydrocephalus [6, 7]. The HCQ-P was developed in parallel with the HOQ and was based on several parent focus groups, followed by rigorous item reduction process, which was also based on parental responses. Our study also has limitations, however. Participation in this study was voluntary and required attendance at a neurosurgery out-patient clinic visit. There are unavoidable biases involved with this recruitment process that we cannot account for. Some of the questions answered by ETV parents were worded slightly differently than those of shunt parents in order to make them more relevant (Q4, Q8, and Q9). These were the same questions for which responses differed the most between ETV and shunt parents. Therefore, it is possible that some of the differences we observed in responses might be attributable to parents interpreting these questions differently in a systematic way.

The concept of parental concern is one that is familiar to any pediatric neurosurgeon. Because most clinical decisions in pediatrics involve parents, their concern can impact significantly on the medical management of their child. The concept of parental concern is not simple, however. The literature suggests that parental adjustment to a serious illness in a child, such as hydrocephalus, involves a complex interaction of various concepts, including several cognitive and emotional processes, sorrow, and illness-related uncertainty [1]. There are certain positive features; what might be termed "appropriate" concern. This ensures that the child receives timely medical care or other forms of support when legitimately needed, for example. Parental concern does, in most cases, reflect true child pathology and so must be appreciated [3]. There are also negative features, however, which might be termed "inappropriate" or excessive concern. This has been linked with worse long-term outcome in some pediatric patients but the causal association is still debated: does worse child outcome lead to more parental concern, or can more parental concern adversely impact a child's outcome [8–10]? At the other extreme of the spectrum is the possibility of inappropriate lack of concern, which could deprive the child of much needed care and support. This would be important in hydrocephalus, since the level of parental concern will often impact the decision of when a common symptom, like headache, results in an urgent hospital visit.

Our results are open to interpretation. The lower levels of concern expressed by ETV parents could very well be viewed as a positive and healthy contribution of ETV to the lives of children and their families. Worrying less about the need for future surgery or complications can allow the family to assume a more normalized existence. This could be valuable information to provide to families when they are deciding between ETV and shunt for their child. On the other hand, is the lower concern actually indicative of an inappropriate lack of concern? That is, are ETV parents not fully appreciative of the fact that ETVs, just like shunts, can block and need repeat surgery [2]? It is the routine practice of all neurosurgeons at our institutions to inform and routinely remind parents about this possibility. Nevertheless, parental concern about this issue is much less than for the parents of children with shunt. It is true that the ETV patients spent fewer days in hospital for hydrocephalus complication management. Therefore, it might be legitimately expected that ETV parents would have less concern about this issue. The lower concern among ETV parents remains, however, even after we adjusted for the child's previous history of neurosurgical complications. This should, therefore, be a continual focus of education of parents, in order to ensure that signs and symptoms of raised intracranial pressure, if they do occur, are dealt with in an appropriately serious and expeditious manner.

Conclusions

Parents of children who have had ETV experience less concern than those who have had shunt, and this relates almost exclusively to less concern about hydrocephalus treatment complications. While this could be interpreted as a beneficial aspect of ETV treatment, it remains important for neurosurgeons to ensure that parents are not being overly complacent about the possibility of ETV failure requiring urgent treatment.

Funding Funding for this study was provided by Sick Kids Foundation, Canadian Institutes of Health Research (Institute for Human Development, Child and Youth Health), IWK Health Centre Foundation, and the Jonathan Lawson Endowment for Spina Bifida.

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