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Quality of life in obstructive hydrocephalus: endoscopic third ventriculostomy compared to cerebrospinal fluid shunt

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Abstract
Purpose In the current literature, there are essentially no comparisons of quality of life (QOL) outcome after endoscopic third ventriculostomy (ETV) and shunt in childhood hydrocephalus. Our objective was to compare QOL in children with obstructive hydrocephalus, treated with either ETV or shunt.
Methods A cross-sectional survey was conducted at SickKids, Toronto of children between ages five and 18 years, with obstructive hydrocephalus due to aqueductal obstruction and no other brain abnormalities. Measures of QOL were the Hydrocephalus Outcome Questionnaire and the Health Utilities Index Mark 3. A subset of patients was given the Wechsler Intelligence Scales for Children (WISC-IV).
Results A total of 47 of 59 (80%) eligible patients participated (24 had ETV as primary treatment, 23 had shunt as primary treatment), with a mean age of 12.1 years (standard deviation 3.9) at assessment. The ETV group was older at initial surgery ($p<0.001$) and had larger ventricle size at last follow-up ($p=0.047$). In all QOL measures, there were no significant differences between the ETV group and shunt group (all $p\geq 0.09$). Treatment failure, hydrocephalus complications, and the presence of a functioning ETV at assessment were not associated with QOL differences. Among the 11 children (six ETV, five shunt) who were given the WISC-IV, there were no significant differences between the scores of the ETV group and shunt group (all $p\geq 0.11$).
Conclusions This is the first study to provide a meaningful comparison of QOL after ETV and shunt in children. These preliminary results suggest that there is no obvious difference in QOL after ETV and shunt.

Keywords Hydrocephalus · Pediatric · Health outcome · Quality of life

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Introduction

The relative efficacy of endoscopic third ventriculostomy (ETV) and cerebrospinal fluid (CSF) shunt in the treatment of childhood hydrocephalus remains a matter of ongoing debate. Few studies have attempted to provide a direct comparison of these treatments [2, 7, 18]. None have yet provided a meaningful comparative analysis of quality of life (QOL), even though patients with hydrocephalus view QOL as the most important outcome of their treatment [8]. There are many reasons to believe that QOL could be appreciably different with these two treatments, including the effects of surgical injury to deep brain structures, surgical complications (e.g., obstruction, infection, overdrainage), and differences in ventricle size.

The goal of our study was to compare QOL in a relatively homogeneous group of children with obstructive hydrocephalus treated with either ETV or shunt using the validated Hydrocephalus Outcome Questionnaire (HOQ) [10, 11] as our primary outcome measure. Our previous studies on QOL have involved large samples of children, covering the broad spectrum of pediatric hydrocephalus

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[13, 14]. For this study, however, we limited our analysis to a highly select group: children whose hydrocephalus was caused by discrete midbrain pathology, without evidence of any other anatomical brain abnormalities.

Clinical materials and methods

A review of the neurosurgery database at SickKids, Toronto was conducted to identify patients meeting the following inclusion criteria: between five and 18 years of age with a diagnosis of obstructive hydrocephalus, which was initially treated at least 6 months earlier, caused by discrete midbrain pathology (primary congenital aqueductal stenosis or extrinsic aqueduct obstruction due to tectal glioma or other localized anatomical lesion), with no history of any other neurosurgical interventions other than for hydrocephalus treatment (e.g., no tumor resection), no history of brain infection or hemorrhage prior to treatment, no chemotherapy or radiotherapy, and with no evidence of other anatomical brain abnormality. These patients were approached either during their regularly scheduled neurosurgery out-patient clinic visit [13] or by mailed surveys, supplemented with direct telephone contact. Participants were compensated for their time with a small monetary gift. All patients in this study had received their entire neurosurgical care at SickKids.

The medical charts and brain imaging were then reviewed by the research team to extract further information about the child's medical history. In order to capture both the frequency and the severity of surgical complications, the *length of hospital admission* was recorded for treatment of CSF infection, shunt/ETV obstruction, CSF overdrainage, and for observation only [13]. This represented the sum of all hospital admissions required for each complication over the child's lifetime, since this was felt to more accurately reflect the severity of the complications. For cases in which multiple complications occurred during the same admission, the length of stay was attributed to the initial problem that brought the child into hospital. Ventricle size was measured using the frontal–occipital horn ratio (FOR), a previously validated linear measure of ventricle size [9, 15]. This was assessed for both the initial (pretreatment) ventricle size and for current (posttreatment) ventricle size.

QOL outcome measures Parents were asked by a trained research assistant to complete the Hydrocephalus Outcome Questionnaire (HOQ) and Health Utilities Index Mark 3 (HUI-3). The HOQ is a reliable and previously validated measure of QOL with scores ranging from 0 (worst QOL) to 1.0 (best QOL) [10, 11]. 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 [10].

The HUI-3 provides utility scores, which are estimates of the general population's relative preference for a given state of health, from 0 ("dead") to 1.0 ("perfect health") [6]. Utility scores can be compared to established population norms [3–5].

Neuropsychological measures A limited number of patients in our study took part in formal neuropsychological testing as part of a study protocol that began in 2006. These patients were selected as part of a convenience sample accrued during clinic visits. As part of a larger battery of tests, these children completed the Wechsler Intelligence Scale for Children-Fourth Edition (WISC-IV), which provided a number of measures including full scale intelligence quotient (IQ), a verbal comprehension index score (hereafter called verbal IQ), and a perceptual reasoning index score (hereafter called nonverbal IQ).

Statistical Analysis Between-group comparisons were performed with independent samples *t* test (for continuous variables) or chi-squared (for proportions). Correlations between continuous variables were calculated using Pearson's correlations. We considered any p values of <0.05 to represent associations of interest, but we recognized our analyses as exploratory in nature.

All analyses were performed using SPSS Advanced Statistics 17.0 (SPSS Inc., Chicago, IL, USA). This protocol was approved by the SickKids Research Ethics Board (number 1000005376).

Results

From a total of 59 patients who were identified to have met the inclusion criteria, 47 participated in the study (80%). Detailed patient characteristics are listed in Table 1. The ETV group was older at initial surgery ($p<0.001$), had shorter duration of hydrocephalus ($p=0.002$), had fewer cases of primary aqueductal stenosis ($p<0.001$), had fewer cases requiring repeat surgery ($p=0.03$), had larger ventricle size at last follow-up ($p=0.047$), spent fewer days in hospital for CSF obstruction ($p=0.005$), and spent fewer days per year in hospital for any hydrocephalus-related complications ($p=0.02$). Five patients had a history of premature birth: three in the shunt group (born at 30, 31, and 34 weeks) and two in the ETV group (born at 32 and 34 weeks). Amongst the seven ETV patients who failed their initial treatment, all went on to have a CSF shunt and were, therefore, shunted at the time of assessment. One patient initially treated with shunt had a functioning ETV at the time of assessment.

Table 1 Patient characteristics

Variable	ETV as first treatment (N=24)	Shunt as first treatment (N=23)	p value
Current age (years)	12.3 (4.0)	12.0 (4.0)	0.77
Age at first surgery (months)	57.3 (57.7)	8.2 (17.4)	≤0.001
Duration of hydrocephalus (years)	6.7 (4.7)	11.0 (3.6)	0.002
Primary aqueductal stenosis (versus extrinsic aqueduct compression)	13 (54%)	23 (100%)	≤0.001
Initial ventricular size (FOR)	0.58 (0.09)	0.62 (0.09)	0.17
Current ventricular size (FOR)	0.48 (0.10)	0.43 (0.09)	0.047
Number who required at least one further hydrocephalus-related surgery	7 (29%)	14 (61%)	0.03
Number of days spent in hospital for initial admission	7.8 (6.0)	8.5 (6.4)	0.71
Number of days spent in hospital for hydrocephalus complications			
CSF infection	0	2.9 (10.2)	0.18
CSF obstruction	1.7 (4.3)	7.5 (8.3)	0.005
CSF overdrainage	0.1 (0.4)	0.1 (0.6)	0.78
Observation only	0.7 (1.6)	1.5 (2.4)	0.85
Number of days per year spent in hospital for hydrocephalus complications (since the initial treatment of hydrocephalus)	0.3 (0.5)	1.2 (1.6)	0.02

CSF cerebrospinal fluid, *ETV* endoscopic third ventriculostomy, *FOR* frontal–occipital horn ratio

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

QOL measures All QOL outcome measures were slightly lower in the 24 ETV patients compared to the 23 shunt patients, although none was significantly different (see Table 2). We tested whether these QOL outcome measures were different between:

a) the 21 patients who failed their initial treatment (14 shunts and seven ETV's) and the 26 who did not (nine shunts and 17 ETV's) and there were no significant differences (all p values were>0.46);
b) the 18 patients who had functioning ETV's at the time of assessment and the 29 patients with functioning shunts at the time of assessment and there were no significant differences (all p values>0.38); and
c) the 11 patients with extrinsic aqueductal obstruction and the 36 patients with primary aqueductal stenosis and there were no significant differences (all p values>0.50).

We tested Pearson's correlations between all QOL outcome measures and number of days per year spent in hospital for hydrocephalus treatment, but none was significant (all p values>0.41).

We tested Pearson's correlations between the QOL outcome measures and current ventricle size (FOR), and found a significant negative correlation with HOQ Physical Score (−0.37, p=0.02) and HUI-3 utility score (−0.37, p=0.01). All other correlations were not significant (all p values>0.06).

Neuropsychological measures The characteristics of the 11 patients who participated in neuropsychological testing are shown in Table 3. The ETV group was slightly younger at assessment (p=0.04) and had fewer cases of primary aqueductal stenosis (p=0.02). All IQ scores were slightly lower in the ETV group, but none was significantly different (see Table 4).

Table 2 Patient quality of life

Outcome variable	ETV as first treatment (N=24)	Shunt as first treatment (N=23)	p value
HOQ Overall Score	0.81 (0.19)	0.85 (0.16)	0.49
HOQ Physical Score	0.88 (0.16)	0.89 (0.15)	0.72
HOQ Cognitive Score	0.72 (0.26)	0.79 (0.25)	0.32
HOQ Social–Emotional Score	0.79 (0.21)	0.86 (0.13)	0.19
HUI-3 utility score	0.82 (0.25)	0.91 (0.18)	0.09

CSF cerebrospinal fluid, *ETV* endoscopic third ventriculostomy, *HOQ* Hydrocephalus Outcome Questionnaire, *HUI-3* Health Utilities Index Mark 3

Values presented are mean (standard deviation)

Table 3 Characteristics of patients who underwent neuropsychological testing

Variable	ETV as first treatment (*N*=6)	Shunt as first treatment (*N*=5)	*p* value
Current age (years)	9.3 (3.0)	13.3 (2.4)	0.04
Age at first surgery (months)	52.6 (54.2)	3.5 (1.7)	0.08
Primary aqueductal stenosis (versus secondary extrinsic compression)	2 (33%)	5 (100%)	0.02
Initial ventricular size (FOR)	0.66 (0.09)	0.67 (0.09)	0.87
Current ventricular size (FOR)	0.53 (0.10)	0.47 (0.11)	0.34
Number who required at least one further hydrocephalus-related surgery	2 (33%)	4 (80%)	0.12
Number of days spent in hospital for initial admission	7.7 (3.4)	8.4 (4.2)	0.76
Number of days spent in hospital for hydrocephalus complications:			
CSF infection	0	4.6 (10.3)	0.30
CSF obstruction	1.8 (3.0)	6.2 (4.6)	0.09
CSF overdrainage	0	0	
Observation only	0	0	
Number of days per year spent in hospital for hydrocephalus complications (since the initial treatment of hydrocephalus)	0.3 (0.5)	1.1 (1.4)	0.22

CSF cerebrospinal fluid, *ETV* endoscopic third ventriculostomy, *FOR* frontal–occipital horn ratio

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

None of the IQ scores were significantly different when we compared: (a) any treatment failure (*N*=6) to no treatment failure (*N*=5); and (b) functioning ETV at assessment (*N*=5) to functioning shunt at assessment (*N*=6) (all p values≥ 0.58). Current ventricle size (FOR) was negatively correlated with all WISC-IV IQ scales (full scale IQ, verbal IQ, and nonverbal IQ), but none of these correlations was statistically significant (all p values≥0.37).

Discussion

Our preliminary results suggest that there is no obvious difference in QOL between ETV and shunt. We examined other possible confounders for association with outcome, including treatment failure, overall complications, and ventricle size. This revealed that larger ventricles appeared to be significantly correlated with poorer HOQ Physical Score and HUI-3 utility score.

Strengths of our study include the strict inclusion criteria, which created a homogeneous population of patients in whom other brain abnormalities were excluded. This resulted in a high-functioning sample of children, with relatively high QOL scores (the mean HOQ Overall Health Score was>0.80, compared to our previous large scale studies in which the mean sample scores were 0.65 [14] and 0.68 [13]). We used measures of QOL with proven reliability and validity and, in a subset of our patients, we used well-accepted, standardized IQ measures.

We acknowledge methodological limitations in our study, as well. Our sample size was limited, particularly for neuropsychological outcomes, thereby reducing the statistical power of our analysis. Regardless, statistical significance was demonstrated in at least some measures and the sample size had 80% power to detect a difference in QOL of at least 0.15 points on the HOQ scale (which we have previously shown to be a clinically important difference [12]). The sample size was too small, however, to perform multivariable regression analysis, which would have allowed us to assess and adjust for several variables simultaneously. Our response rate was 80% and so, our sample might be biased and not wholly representative of this group of patients. Furthermore, despite our strict inclusion

Table 4 Neuropsychological test results

Outcome variable	ETV as first treatment (*N*=6)	Shunt as first treatment (*N*=5)	*p* value
WISC-IV full scale IQ	92 (20)	107 (18)	0.23
WISC-IV verbal IQ	98 (12)	111 (12)	0.11
WISC-IV nonverbal IQ	91 (17)	102 (10)	0.23

ETV endoscopic third ventriculostomy, *WISC-IV* Wechsler Intelligence Scales for Children, 4th Edition; *IQ* intelligence quotient

Values presented are mean (standard deviation) criteria, the ETV and shunt groups were still systematically different. For example, the ETV patients were older at first surgery and all patients with extrinsic aqueduct obstruction were exclusively in the ETV group, representing a selection bias amongst the treating surgeons. This makes it virtually impossible to separate out these effects from those of actual treatment on outcome. As well, the shunt group had a longer duration of hydrocephalus and more hydrocephalus complications, which we have previously shown to be associated with worse QOL [13, 14]. If anything, however, this should have favored the ETV group. Our study did not include comparison of pretreatment QOL and cognitive functioning, which might have revealed further systematic differences between the ETV and shunt groups. This would be virtually impossible to do, however, since many of the shunted patients were treated as young infants and there is no accepted way of measuring QOL in such young patients.

Although we are not aware of any previous work that has compared QOL after ETV and shunt, there is some limited work in assessing intellectual functioning after ETV. For example, Burtscher et al. showed improvement in cognitive functioning after ETV in six adults with late onset aqueductal stenosis, but there was no shunt comparison group [1]. Takahashi reported that, among infants with aqueductal stenosis, long-term intellectual outcomes were superior with shunt compared to ETV in those who present with severely expanded ventricles and transependymal edema [17].

We fully recognize that, given the limitations of our study, our results are to be regarded as only preliminary. At a minimum, however, our results suggest the need for further study in this area. The International Infant Hydrocephalus Study, currently the only randomized comparison of ETV and shunt, will be examining long-term QOL and neuropsychological outcomes in infants with obstructive hydrocephalus as its primary measures [16]. This sample should be very homogeneous and, therefore, the effect of confounding should be minimal. The results, however, are several years away. In the meantime, our group is planning a more detailed assessment of all children at our institution with obstructive hydrocephalus related to discrete midbrain pathology and subjecting them to detailed assessments of QOL, neuropsychological functioning, diffusion tensor imaging, and volumetric assessment of ventricle size. A study of this nature might be able to provide a reasonable physiological explanation for any differences in outcome observed between ETV and shunt.

Conclusions

Our study in children with obstructive hydrocephalus from discrete midbrain pathology suggests that QOL and intellectual outcomes following ETV and shunt are not substantially different. These are preliminary results, however, in a limited sample of patients. Important confounding effects cannot be excluded. Larger and more detailed studies are needed.

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