Efficacy of endoscopic third ventriculostomy in the management of hydrocephalus in children under 2 years of age: Experience from a tertiary institution in Nigeria

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Abstract

Background: The management of hydrocephalus in developing countries is challenging. Hydrocephalus is a common childhood disorder in developing countries in particular and its management is quite challenging. Ventriculoperitoneal (VP) shunt is associated with high failure rates and complications. Endoscopic third ventriculostomy (ETV) with potentially lower complication rate could improve care and reduce cost of management of hydrocephalus.

Objective: The aim of this study was to evaluate the efficacy (success rate) of ETV in children ≤2 years and to find out factors that may be responsible for good outcome of ETV.

Methods: This prospective observational study was conducted at Lagos University Teaching Hospital, Lagos. Nigeria. All consecutive children ≤2 years of age with hydrocephalus were recruited into the study. Relevant demographic and clinical data documented. All cases had ETV and were followed up to document 6 months outcome.

Results: A total of 34 patients (M: F ratio 1.1:1) were recruited over a 2-year period. Age, sex, presumed aetiology and image findings were not statistically significant in influencing outcome of ETV. Good outcome (defined as uneventful postoperative period, not requiring repeat ETV or VP shunt) was documented in 26 (73.5%). A total of 8 (26.5%) experienced poor outcome. Complication occurred in 2 (5.9%) as follows: Wound infection 1 (2.9%) and ventriculitis 1 (2.9%). Aetiology was divided into non post-infective hydrocephalus-20 (58.8%), post-infective hydrocephalus-5 (14.7%) and post-myelomeningocoele repair-9 (26.5%).

Conclusions: This study shows that ETV success rate is high in the management of hydrocephalus in children ≤2 years in our clinical practice. Regardless of the clinical diagnosis, where the facilities are available, children with hydrocephalus will benefit from ETV irrespective of the age and aetiology in sub Saharan Africa.

Key words: Children, endoscopic third ventriculostomy, hydrocephalus, ventriculoperitoneal shunt

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Introduction

Hydrocephalus is defined as an active distension of the ventricular system of the brain resulting from inadequate passage of cerebrospinal fluid (CSF) from its point of production within the cerebral ventricles to its point of absorption into the systemic circulation.[1,2] Globally, the burden of hydrocephalus in the paediatric population is high, with prevalence rate of 1.2/1000 children.[3] The rates vary significantly across populations, but hydrocephalus may account for up to 50% of cases treated by neurosurgeons in the United Kingdom.[4] There are no local data regarding

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the hospital frequency or population-based prevalence data for hydrocephalus.

The impact of hydrocephalus on the patient, caregivers and the healthcare system is enormous. Hydrocephalus, untreated, can result in delay of developmental milestones, blindness, learning difficulties and eventually death. The economic impact of hydrocephalus runs into millions of US dollars in sub-Saharan Africa.[5]

The recommended surgical treatment options for hydrocephalus are ventriculoperitoneal (VP) shunt and more recently, endoscopic third ventriculostomy (ETV). VP shunts are traditionally associated with high complication rates (approaching 40%) including infection, shunt extrusion, shunt malfunction, and breakage.[6,7] This is in contrast to ETV, which avoids foreign body insertion, is a shorter procedure with expertise, and may be more cost effective for patients and hospital systems.[6] Complication rates reported for ETV are <10% in various series.[8] Specifically in our practice environment, VP shunts have been associated with an overall complication rate of 25.8%, and an infection rate of 19.5%.[9] This provides a justification for exploring an alternative procedure such as ETV with potentially lower complication rates, and by extension, shorter hospital stay and lower direct costs to patients undergoing VP shunts. There are no data from our practice environment describing outcomes from ETV, although such information is necessary to guide clinical practice and provide a basis for any modification in current treatment approach for hydrocephalus for neurosurgeons practicing in developing countries such as ours.

This observational study was designed to describe, using a prospective approach, the outcome following ETV at our practice site in Lagos, Nigeria. The study aimed to document the medium term outcome (at 6 months) in children aged ≤2 years, with the primary outcome measure being a survival without the need for any additional intervention. In addition, the study explored the factors associated with the desired outcome. Specific factors explored were: The effect of age (in months), aetiology of hydrocephalus, and imaging findings with respect to type of hydrocephalus (tri-ventricular or panventricular).

Methods

The study protocol was approved by the Ethics Committee of the Lagos University Teaching Hospital, Lagos State, Nigeria (LUTH). LUTH is a tertiary hospital with a full complement of surgical and medical specialties, including, at present, three neurosurgeons. The hospital rate of hydrocephalus at the time of the study was, on average 60 cases annually (and 40% of all operated neurosurgical cases). All cases of hydrocephalus are managed by the neurosurgical unit. Prior to 2005, all paediatric cases of hydrocephalus who underwent surgery were treated with VP shunt. However, following acquisition of expertise for ETV, surgical intervention of choice became ETV for the majority of cases, with VP shunt restricted primarily to cases with failed ETV.

Study design and recruitment procedure

The study employed a prospective observational design, recruiting all consecutively referred cases of hydrocephalus aged ≤2 years in whom consent for surgical intervention was obtained from the parents/guardians. Patient recruitment was conducted over a 2-year period between 2009 and 2011. The only exclusion criteria were age >2 years or nonconsent. However, all cases consented to participate in the study. Participants were recruited from the paediatric surgery clinic, emergency unit, neurosurgery out-patient clinic and in-patient service.

Clinical evaluation and data collection

Clinical data were obtained from parents/guardians and case records. Baseline data included patient demographics, parent/guardians personal data (educational level, socioeconomic status), medical history, birth history and maternal gestational history for patient's birth. Clinical history relevant for etiologic determination and physical findings were documented.

All cases underwent radiological evaluation (trans-fontanelle ultrasound scan (TFUSS) and/or brain computerized tomographic scan) at presentation. Hydrocephalus was defined as the presence of dilated ventricles on TFUSS[10,11] or brain computed tomography (CT) scans.[11] Furthermore, hydrocephalus was subdivided into tri-ventricular (obstructive) (in the presence of dilated lateral and third ventricles only) or panventricular (communicating) (with involvement of the fourth ventricle).[3]

Based on clinical data, the aetiology of hydrocephalus was presumed to be one of the following: Post-meningomyelocele repair (hydrocephalus developing after meningomyelocele repair), post-infective hydrocephalus (PIH) (patients with history of febrile illness indicative of a central nervous system infection preceding onset of hydrocephalus), and non post-infective hydrocephalus (NPIH) (not post-surgical, not post-infectious, overt aetiology such as aqueductal stenosis, tumour in the third or fourth ventricle or covert aetiology, e.g. congenital hydrocephalus).

Surgical procedure

Each patient is usually supine on the operating table with the head in a neutral position[12] or turned to the left. Minicraniotomy is done at the lateral angle of the open anterior fontanel on the right.

Following the craniotomy, the dura was incised and coagulated to control bleeding and prevent blood dripping to the ventricles. A brain cannula was then used to make a trajectory into the ventricle. The trajectory of the endoscope is slightly medial and oriented in line with the external auditory meatus in a posterior direction. This trajectory yields towards the foramen of Monro.[13] The endoscope (2.8 mm flexible Karl Storz [Tuttlingen, Germany] Neuroendoscope 11282BN1) was then introduced and advanced until the lateral ventricle was seen. Further manipulation include following the choroid plexus anteriorly and medially until the foramen of Monroe was entered to gain access to the third ventricle. Ventriculostomy was made with the bugby wire at the midpoint between the infundibula recess anteriorly and the two mammillary bodies posteriorly.

All cases received routine antibiotic (50 mg/kg body weight of ceftriaxone) preoperatively and continued for the next 5 days after surgery.

Postoperative follow-up

All cases were followed up for 6 months postoperatively. To facilitate this, initial post-ETV hospitalization was scheduled routinely for a period of 2 weeks, followed by outpatient evaluations at 2 weekly intervals for the initial 3 months, and then every month for the final 3 months. To limit loss to follow-up, contact details including telephone and home addresses, and follow-up via the unit social worker were also incorporated.

During the postoperative follow-up period, the following assessments were conducted: Documentation of complications based on clinical and laboratory assessments (e.g. surgical site infection, presence of clinical symptoms or signs of raised intracranial pressure) and head circumference measurements (to assess any increase in circumference indicative of failed ETV).

Data analysis

All data collected were documented using a standard proforma. Primary outcome was ETV success rate at 6 months (regarded as good if patient was alive at 6 months and had no indication for repeat intervention and poor if otherwise, i.e. death or need for repeat ETV or VP shunt). Secondary outcome measures were the complication rate following ETV (including infection rate, repeat procedure rate, case fatality rate). All rates are reported as total number (%). Numerical data are presented as mean ± standard deviation. Continuous data are presented as proportions (%). Intergroup comparisons (good versus poor outcome categories) of variables (age, etiology and type of hydrocephalus) were made using ANOVA (for age) and Chi-squrae test (for etiology and type). A $P < 0.05$ was regarded as statistically significant.

Results

A total of 34 patients below the age of 2 years were recruited during the study period, comprised of 16 (47.1%) male and 18 (52.9%) female, giving a male female ratio of 1:1.1. The age range was 0-14 months (mean: 4.32 months overall, 3.62 in males and 4.94 in females).

Imaging findings showed that 85.3% (29) of patients had a tri-ventricular hydrocephalus whereas 14.7% (5) had a pan ventricular hydrocephalus. The presumed aetiology was post-meningomyelocoele repair in 9 (26.5%), PIH in 5 (14.7%) and non post-infective in 20 (58.8%).

Of the 34 cases recruited, 26 (73.5%) experienced a good outcome at 6 months post-ETV as shown in Table 1 in contrast to 8 (26.5%) who had a poor outcome as defined in this study. Of the 8 with poor outcome, the reasons were as follows: Failed ETV requiring repeat ETV (2), failed ETV requiring VP shunt (2), deaths within 3 months post-ETV (2), discharged against medical advice or lost to follow-up (2). Postoperative complications within the 1st week postoperatively were documented in 2 (5.9%) of all cases as follows: Wound infection in 1 (2.9%) and ventriculitis in 1 (2.9%). Thus, post-ETV infection rate was 5.9% (2/34).

To explore the relationship between age and outcome at 6 months, the proportion of good versus poor outcome in cases aged 6 months and below and those above 6 months were compared [Table 1]. Good outcome was recorded in 19/26 (73.1%) of children aged 0-6 months compared to 7/8 (87.5%) of those above 6 months ($P = 0.40$). The mean age of cases (months) with good outcome was 4.8 ± 3.9 compared to those with poor outcome, which was 2.8 ± 3.2 ($P = 0.68$).

Table 1: Comparison of outcomes based on age, presumed aetiology and imaging findings

Variable	Good outcome (%)	Poor outcome (%)	*P*
Age (%)			
Age≤6 months (26, 76.5)	19 (73.10)	7 (89.50)	0.400
Age>6 months (8, 23.5)	7 (26.9)	1 (12.5)	
Presumed aetiology (%)			
Postmeningomyelocele surgery (9, 26.5)	9 (34.6)	0 (0.0)	P=0.136, χ^2=3.988
Postinfectious (5, 14.7)	3 (11.5)	2 (25.0)	
Nonpostinfective, nonpostsurgical (20, 58.8)	14 (53.8)	6 (75.0)	
Imaging findings (%)			
Trilateral hydrocephalus (29, 85.3)	23 (88.5)	6 (75.0)	P=0.347, χ^2=0.884
Panventricular hydrocephalus (5, 14.7)	3 (11.5)	2 (25.0)	

The outcome based on the presumed aetiology of hydrocephalus is shown in Table 1. Good outcome was recorded in 9/9 (100%) with post-meningomyelocole hydrocephalus, 3/5 (60%) with post-infectious hydrocephalus, and 14/20 (70%) with noninfectious hydrocephalus. The presumed aetiology is not statistically significant in affecting outcome at 6 months ($P = 0.14$, $\chi^2 = 4.0$).

Out of the total 34 patients, 29 had tri-ventricular hydrocephalus on imaging while 5 had panventricular hydrocephalus. Of the 29 with tri-ventricular hydrocephalus, 24 had a good outcome at 6 months post-ETV giving a good outcome rate of 86%. Good outcome rate was documented in 3/5 (60%) for panventricular hydrocephalus [Table 1]. The difference in outcomes based on the image findings was not statistically significant ($P = 0.33$, $\chi^2 = 0.88$).

Discussion

The study provides data based on the experience with ETV in infants with hydrocephalus in our tertiary, developing country's neurosurgical practice. The main finding from this study is that ETV success rate for hydrocephalus of diverse aetiologies in this setting is high (73.5%). In addition, a trend of better success rate in tri-ventricular hydrocephalus compared to panventricular hydrocephalus was documented although this was not significant.

In a similar study done by Sufiano *et al*. on children <2 years, he recorded a success rate of 75.5% for primary ETV.[14] Most studies both in developing and developed nations had reported an ETV success within the range of 70-80%.[15-18]

The standard imaging modality for diagnosis and classification of hydrocephalus is a brain CT scan. Use of MRI does not add any advantage but rather more expensive. In this study, due to the fact that many patients were poor and health care is paid for the patients or relatives out of pocket, a TFUSS with a print out of the study was used as an alternative for patients that could not afford a brain CT scan. The scan printout showing the ventricles helps in classifying the type of hydrocephalus based on the ventricles that were dilated. There was no need for further imaging postoperatively as serial head circumference and relief of clinical symptoms of hydrocephalus would point to success or otherwise of the procedure.[19]

The success rate of VP shunt as treatment for childhood hydrocephalus is in the range of 50-60%, which is lower than success rate of ETV.[20] Complication rate in VP shunt is in the range of 23% to 25.8% as compared to a lower rate of 6-8% in ETV.[9,21,22] Hence ETV is more beneficial to hydrocephalic children even with the added advantage of patient not requiring foreign body of shunt.

It is the routine in the unit to use endoscope to view the site of previous ventriculostomy in failed ETV patients. All patients who failed ETV were therefore re-scoped and still reported as failed ETV. They were not re-included in the study even if they had good outcome at redo ETV. The ventriculostomy in the three patients that had redo ETV were noted to be closed with fibrous adhesions. They had repeat ventriculostomy and had good outcome but were not included in the success group. We cannot say specifically the reason for this insufficiency.

The most severe intraoperative complication of ETV is rupture of basilar artery. With attention to appropriate landmarks, this can be avoided. We did not record any significant intraoperative complications except for some bleeding from ependymal vessels that stopped spontaneously with irrigation. Training and experience of the surgeon will ensure that complication rates are low. Moreover, with experience complications will be appropriately handled without panic, which might be the case in surgeons with little experience. Ersahin and Arslan agreed that training, experience and meticulous technique would reduce complication rates in ETV.[23]

Postoperative complications encountered were not different from those reported from other countries.[15] Postoperative infection was seen in 1 (2.9%) in the study. We have a protocol of giving antibiotic for a minimum of 5 days due to the prevailing tropical circumstances in the environment. The basics for this practice is anecdotal. It was a fall out of when there was a high rate of postoperative infection seen following VP shunt. This is however yet to proven scientifically as documents in the literature suggest no benefit of prophylactic use of antibiotics beyond 24 h.[24] This is possibly the reason the infection rate is low. Less complications were seen in ETV when compared to VP shunt because the possibility of wound breakdown and extrusion of shunt in malnourished patients were eliminated and patients had shorter hospital stay and were less likely to acquire hospital-borne infections.[6,15] There was no incidence of postoperative CSF fistula in this study though it is a possible complication.[25]

The potential determinants of outcome explored in this study were age, clinical diagnosis and image findings. Of these none showed any significant association with outcome. This is similar to the finds of Sufianov *et al*. and Lipina *et al*.[14,26] However In multicentre study done in a Canada, the correlation between age and efficacy is at variance with this study.[27] The Canadian study found age to be a determinant factor in the success of ETV. The sample population in Canadian study included children with brain tumour and a multicentre study. These are possible reasons why the outcome is different.

In other studies, patients with aqueductal stenosis did better with ETV and this study result showed patients with

obstructive hydrocephalus had better outcome from ETV as well.[28] This study corroborate similar trend of higher success in obstructive hydrocephalus though not statistically significant. This is because ETV creates an alternative physiological pathway to the blocked aqueduct of Sylvius.

Postoperative imaging to assess the efficacy of ETV is not necessary.[15] This is due to the fact that the dilated ventricle will not revert back to its prehydrocephalic state quickly but if the occipitofrontal circumference is not increasing and the clinical features of active hydrocephalus are absent, the procedure is regarded as successful and the outcome is said to be good.

Two of the patients in the study died within the first 3 months of ETV. They died as a result of delay in accessing medical care and on-going debility of the hydrocephalus.

This study shows that the result of the efficacy of ETV is similar to what was obtained in other studies internationally.[14]

Although our study provides data that was not previously available from our practice setting, it has certain limitations. The small sample size of the study may have limited the ability to demonstrate any significant relationship between the determinants assessed and outcome. Also routine use of antibiotics may be responsible for reduced infection rate. There were no comparator group and no randomization of treatment category. However, the study provides an insight into the performance efficacy of ETV that can serve as a basis for future studies.

Conclusion

This study shows that ETV is effective in the management of hydrocephalus in children below the age of 2 years. The complications are fewer than the published rate for conventional shunt procedures.[6] Regardless of the clinical diagnosis, it is worthwhile to do ETV for children with hydrocephalus especially when there are no contraindications such as distorted anatomy and bloody/cloudy CSF.

Recommendation

On the basis of the preliminary data from this study, ETV appears to be a rational and effective alternative to VP shunt in the management of hydrocephalus in infants in our practice setting. Training to expand expertise of neurosurgeons to enable wider application is recommended.

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