

Intraoperative Findings During Neuro-Endoscopy for Hydrocephalus in Tanzania

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Abstract

Background: Endoscopic third ventriculostomy (ETV) can be performed to treat infantile hydrocephalus with variable success.

Methods: We included patients undergoing ETV+/-Choroid plexus cauterization (CPC) in Dar Es Salaam, Tanzania from November 2011-February 2019. Prospective data collection was carried out by the chief surgeon with a total sample size of 497. A separate database from the hydrocephalus nurse provided one-year mortality outcomes from January 2017-December 2018 in 261 patients.

Results: The median age was 138 days; 6%≤1 month, 57% were 1-6 months and 20% were 6-12 months old. The median occipitofrontal circumferences were: 50cm (age≤1 month old); 49cm (1-6 months old); 55cm (6-12 months old). Cortical mantle was thin in 73% and normal in 27%. The aqueduct of sylvius was visualised and closed in 61% and open in 33%. Yellow deposits were seen in 40% and pus in 3%. The CSF was turbid, bloodstained, yellow and clear in 16%, 1%, 3% and 80% respectively. The prepontine cistern was clear in 73%, scarred in 12%, effaced in 11% and not seen in 4%. The tuber cinereum was thin in 85%, thick in 11% and not fenestrated 4%. The choroid plexus was normal in 65% and scarred in 35%. Clinical follow up at 1-month post op was available in 123 (<25%) patients. At 1-month postop the mortality, re-do ETV and VP shunt insertion rates were 8%, 6% and 8% respectively. From the nurse led database, the one-year overall survival was 87%.

Conclusion: ETV+/-CPC is an established treatment option for infantile hydrocephalus in Tanzania.

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Introduction

Historically, infantile hydrocephalus has been associated with a high mortality rate in Tanzania (1–3). In recent years, the Neurosurgical service in Dar Es Salaam has progressed significantly, resulting in improved care for infants with hydrocephalus. Traditionally, insertion of a Ventriculoperitoneal (VP) shunt was the only treatment option available (1,3–5). With the help of CURE international, endoscopic third ventriculostomy (ETV) and choroid plexus cauterisation (CPC) have been successfully introduced as alternative treatment options (6–11). The CURE protocol has been adopted into daily practice to guide decision making (8). This is a treatment algorithm which relies on the assessment of pre-operative imaging and intraoperative endoscopic findings to determine which patients undergo VP shunting, ETV only and ETV-CPC.

CPC has been developed as an adjunct to ETV to increase the success rate of endoscopic surgery (7,8,11–15). An indication for CPC is following ETV when the aqueduct of sylvius is open, however some neurosurgeons may routinely perform CPC in all cases of infantile hydrocephalus to maximise the likelihood of success. CPC can also be carried out without ETV, such as in Hydranencephaly (16). However, if the choroid plexus appears scarred due to infection, then CPC is not performed. The success rate may potentially be dependent on the extent of choroid plexus which is cauterised (11–13,15). Scarring of the prepontine cistern has been identified as a negative predictor of ETV success (14,15,17). As a result, the CURE protocol favours converting to VP shunting when cistern scarring is encountered during endoscopy (8). However, VP shunt insertion may be contra-indicated due to active cerebrospinal fluid (CSF) infection.

In Tanzania, patients often present to neurosurgical services late. This is because there are geographical, economic and educational barriers to accessing neurosurgical care (6). This may lead to intraoperative findings which represent severe disease due to hydrocephalus. Therefore, the goal of this study was to determine the intraoperative findings from neuroendoscopic procedures performed for hydrocephalus in Tanzania.

Methods

A single centre case series including patients undergoing ETV +/- CPC in Dar Es Salaam, Tanzania from November 2011 to February 2019. Prospective data collection was carried out by the chief surgeon with subjective analysis of the intraoperative neuroendoscopic findings. The total sample size was 497 with a male to female ratio of 1.4:1. Patients were categorized by age; 0 to 30 days as ≤ 1 month, 31 to 183 days as 1 – 6 months, 184 to 365 days as 6 – 12 months, 1 – 10 years and older than 10 years. The clinical outcomes within 30 days of surgery were analysed which included; death, redo ETV and insertion of VP shunt. The examples with neuroendoscopic photos were taken after the period of data collection, this is because the recording technology was previously not available. A second database was analysed, which included mortality on patients who underwent ETV +/- CPC from January 2017-December 2018, collected by the hydrocephalus nurse. This was used to generate a 1-year overall survival. The two databases were not amalgamated.

These preliminary results were not compared against the ETV success score as there are confounding factors which are difficult to control in this clinical setting with the resources we had available. Therefore,

we are unable to definitively calculate the ETV success rate in our patient cohort. There are difficulties with follow up, as the patient population covers a wide geographical area, and the parents may not have readily accessible transport or the financial capacity to attend hospital appointments, due to poverty. There are multiple causes for delayed mortality in this patient population, such as malnutrition, diarrhoeal illness, malaria and other infectious diseases, therefore we were unable to conclude if any mortalities were secondary to recurrent hydrocephalus or systemic disease. We have presented a 1-year overall survival with the data which was available, however further resources and improved infrastructure would be needed to reliably determine the ETV success rate.

Results

Preoperative patient demographics

11% of patients had previously undergone VP shunt insertion and of those who had been tested for Human Immunodeficiency Virus (HIV), 7% were seropositive. The age of patients is presented in figure 1 and the demographics of those under 12 months old in table 1.

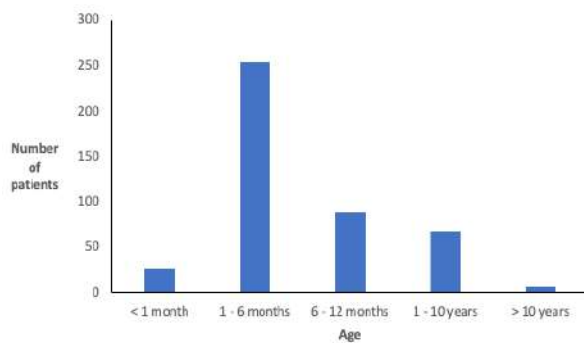


Figure 1: Age of patients undergoing ETV +/- CPC

Intraoperative findings

Figure 2 displays the intraoperative findings during neuroendoscopy. In addition, the septum pellucidum was not intact in 87% and normal in 13%. Figures 3 and 4 display intraoperative photos taken during neuroendoscopy, demonstrating examples of cistern scarring and poor visibility ETV-CPC due to turbid CSF and active infection.

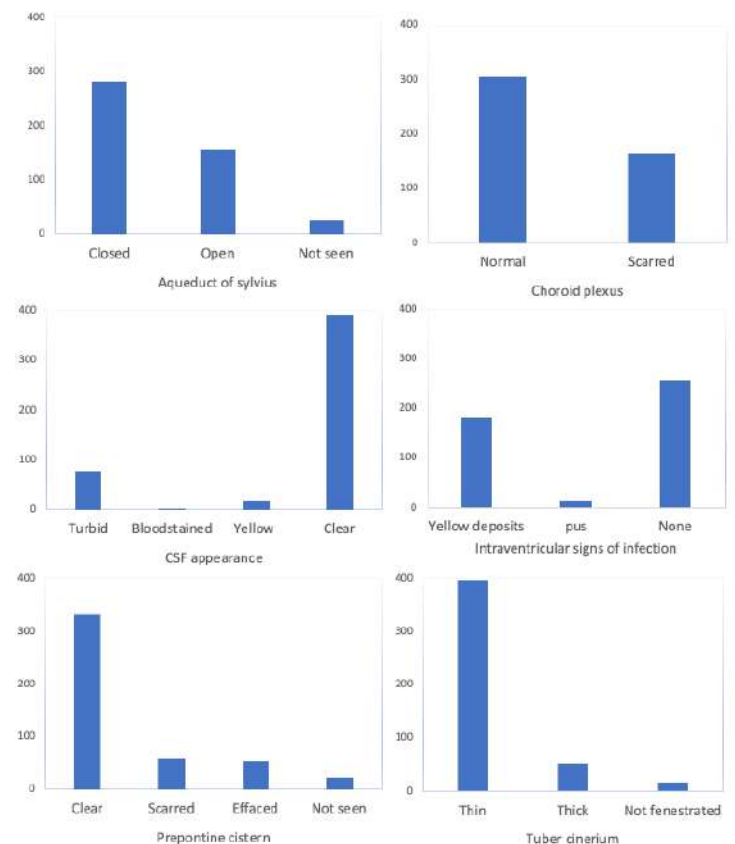


Figure 2: Intraoperative findings during neuroendoscopy

Clinical outcomes

Clinical follow up at 1-month post op was available in 123 (<25%) patients. Within 1-month of surgery the mortality rate was 8%, re-do ETV was 6% and VP shunt insertion was 8%.

The second database identified 261 patients who underwent ETV +/- CPC from January 2017 to December 2018. Within 365 days of surgery, there were 33 (13%) confirmed deaths, however the cause of death was not stated (Figure 5).

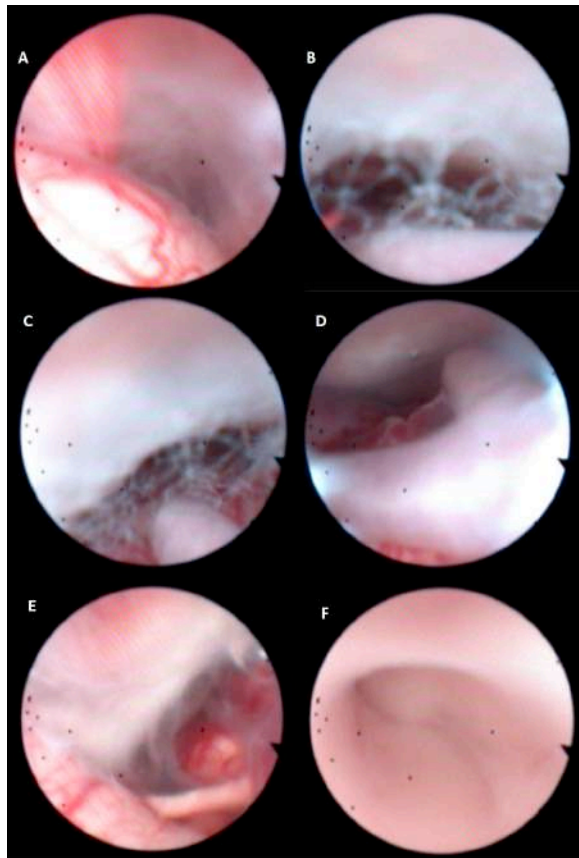


Figure 3: Cistern scarring

A) After fenestration of the tuber cinereum and scarred interpeduncular cistern. B, C) prepontine scarring on the basilar artery. D) Vertebral arteries free floating with good CSF flow through the pre pontine cistern. E) good stoma flow (compare with A). F) sylvian aqueduct closed so CPC not performed.

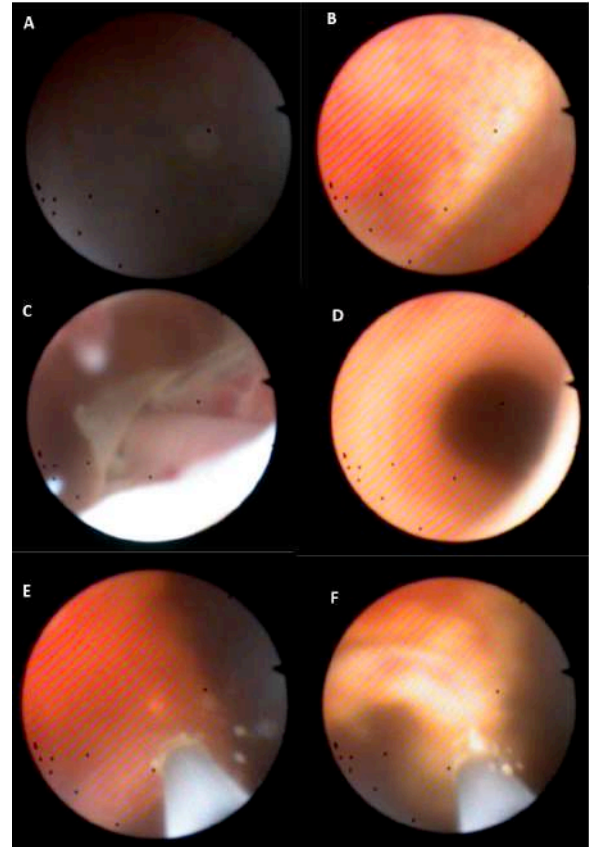


Figure 4: Active CSF infection and poor visibility ETV-CPC

A) View within the lateral ventricle when the CSF is turbid. B) Within the third ventricle, the dorsum sella and pituitary infundibulum with haemosiderin staining secondary to likely infection is visible. C) ETV performed. A wide stoma and good CSF flow was achieved. The prepontine cistern is inspected and no scarring found, liliquist's membrane opened to show the basilar artery. D) Sylvian aqueduct wide open, therefore CPC performed. E, F) CPC performed however unable to conclude the extent of cauterisation due to poor visibility.

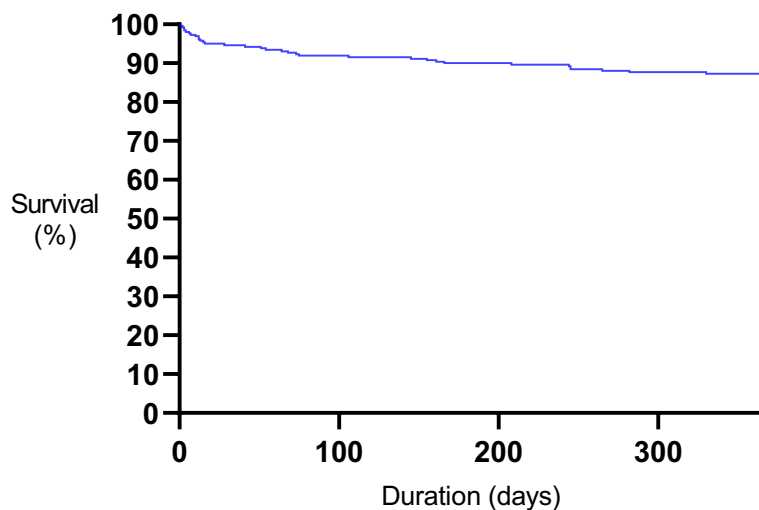


Figure 5: One-year overall survival following ETV +/- CPC from the second database

Table 1: Preoperative demographics

		Age		
		≤ 1 month	1 - 6 months	6 - 12 months
Birth weight (kg)		3.6 (2.0 - 5.0)	3.0 (1.0 - 4.7)	3.0 (1.6 - 4.9)
Weight (kg) at operation		4.4 (3.0 - 10.5)	6.0 (3.0 - 12.0)	8.0 (3.0 - 15.0)
OFC (cm)		50 (37 - 57)	49 (39 - 73)	55 (45 - 70)
Hb (g/dl)		13.5 (8.3 - 18.6)	10.4 (8.8 - 9.4)	10.5 (8.2 - 13.1)
Cortical mantle	Normal	29%	24%	29%
	Thin	71%	76%	71%

Discussion

The introduction of neuroendoscopy for hydrocephalus

The introduction of neuroendoscopy has revolutionised the treatment of hydrocephalus in Sub-Saharan Africa (7,8,11,18) given the high complication rate associated with VP shunts (1,3–5,7,19–22). The overall mortality following paediatric VP shunt insertion reported across Tanzania is 0-21% (1,3–5), however there was a high variability in duration of follow up. The combined shunt and wound infection rates reported are approximately 15-46% (1,3–5).

There are also rare complications following infantile VP shunt insertion which are difficult to manage in a resource limited setting. For example, craniosynostosis secondary to over drainage (20,23,24) and peritoneal dysfunction requiring alternate distal catheter placement (19), such as into the right atrium (25,26) or pleura (27,28).

Given the life-long maintenance required for VP shunts (29), ETV+/-CPC using a flexible endoscope is the first line treatment option in Tanzania. The estimated 6-month success rate of ETV+/-CPC for infantile hydrocephalus is 35-83% (7–9,11–13,16,17,30–37). Despite cistern scarring (8,14,15,17,30,35,38) and poor visibility (30,39,40) being an indication to abandon surgery, ETV +/- CPC can still be performed in select cases (*Figures 3 and 4*).

CSF analysis

In our series, the CSF was turbid in 16% resulting in poor visibility. Nevertheless, the tuber cinereum was fenestrated in 96% of

cases. Currently CSF analysis is not readily available, which means that VP shunts are inserted without confirmed absence of infection. Our study demonstrates that CSF can appear macroscopically clear, however signs suspicious of infection can be seen on neural tissue during endoscopy (*Figure 2*).

Performing preoperative fontanelle taps could determine which patients have active CSF infection and be used when planning a date for surgery. However, fontanelle taps carry a risk of introducing infection. Lumbar punctures are often not performed given the high incidence of myelomeningocele. Sending CSF for microbiological analysis at the time of endoscopy may be useful for patients at high risk of ETV failure and needing a subsequent VP shunt.

Choroid plexus cauterization

CPC has been developed as an adjunct to ETV to improve the success of endoscopic surgery. However, in our series the choroid plexus was scarred in 35% of patients, likely due to previous CSF infection. Furthermore, the sylvian aqueduct was closed in 61% of patients, which could indicate a higher likelihood of success and CPC may not be performed. The CSF was turbid in 16% of cases, while an ETV can be performed through poor visibility, it is difficult to carry out CPC and confirm the extent of cauterisation. Nevertheless, CPC remains a useful procedure for select patients with hydrocephalus.

Predictors of ETV failure

The tuber cinereum was thick in 11% of patients. These cases can be technically difficult and require manoeuvres such as

diathermy to perform the third ventriculostomy, which may increase the risk of bleeding. Cistern scarring was encountered in 12% of cases, which may predict of ETV failure (14,15,17,41). The pre pontine cistern was effaced in 11%, however this may not be a predictor of ETV failure (42) as CSF flow can still occur.

Educating the mothers of infants with hydrocephalus

Young age is a predictor of ETV failure (35–38,43,44). In our series, 63% of patients were aged less than or equal to 6 months old. Therefore, it is important to educate the mothers on the signs and symptoms of hydrocephalus so that patients who require redo ETV or insertion of a VP shunt are brought back to hospital. The mothers of infants with hydrocephalus are often young, living in poverty and have received a low level of education (6). The median age in Tanzania is 18 years old and the rate of poverty has been estimated as high as 65% in regions such as Rukwa and Tabora (45).

We found that amongst Tanzanian mothers of children with spina bifida and hydrocephalus, 59% had obtained a primary school level of education only and 17% had received no school education at all (46). Moreover, the proportion of adult females who are unable to read and write has been estimated at approximately 40% in regions such as Geita and Simiyu (45).

The Association of Spina Bifida and Hydrocephalus Tanzania (ASBAHT), is a non-governmental organisation which aims to improve the quality of life for these children and their families. ASBAHT runs an educational program once a week to help support the mothers.

Macrocephaly and body weight

The mean head circumferences for patients aged <1 month, 1-6 months and 6-12 months were 50cm, 49cm and 55cm respectively. These are all above the 99.9% centile, as per the World Health Organisations growth charts. This likely reflects delayed diagnosis secondary to limited access to healthcare (46) and may contribute to neurodevelopmental delay. In our series, infantile anaemia was common (table 1), likely due to malnutrition. Infantile anaemia can impair cognitive development (47,48) and may potentially contribute to neurodevelopmental disability seen in patients with hydrocephalus. In our series, the body weight at operation was a minimum of 3kg, to avoid the anaesthetic risks associated with surgery on a low weight infant. We observed a wide range of body weight at the time of surgery for the patients younger than 12 months. Low body weight in infancy is likely due to malnourishment. The patients with higher body weight could potentially be due to extreme macrocephaly in some cases. We have observed scalp pressure sores due to cranial weight and mothers who are unable to lift their child so struggle to breast feed them.

Limitations

The study is limited by the lack of long-term clinical outcomes, such as neurodevelopmental assessments. However, due to the severity of CNS infection and delayed treatment of hydrocephalus leading to thinned brain mantle, it is likely that some patients will have cognitive impairment or developmental delay, even if their hydrocephalus is adequately treated (7). In

our series, over 70% of patients younger than 12 months old had thinned cortical mantle pre operatively. Currently, no formal assessment of cognitive development is being carried out. Another limitation is the number of patients with missing clinical data and short follow up in the primary dataset. Therefore, we are likely over estimating the number of patients who remain shunt-free following neuro endoscopic intervention. The causes of death were not available. Given that there are multiple causes of infantile mortality in Tanzania, such as malnutrition (49), malaria (5) and diarrhoeal illness (50), our study may potentially over-estimate the mortality risk due to ETV failure. Our study also lacks information on other complications such as wound infection, CSF leak and seizures. An additional limitation was the subjective nature of interpreting the intraoperative findings. Signs of suspected infection on neural tissue were not confirmed via microbiological diagnosis. Furthermore,

there was no predetermined measurements of the cortical mantle to define thin versus normal. The intraoperative findings were not correlated against the preoperative radiological findings such as size of the fourth ventricle or obstruction at the sylvian aqueduct. The study utilizes two separate databases, however the hospital now prospectively completes one common database on RedCap which will likely lead to high quality data in the future.

Conclusion

The intraoperative findings may reflect delayed diagnosis of hydrocephalus or severity of infection, and could be used to guide decision making with regards to ETV +/- CPC. This study is limited by missing clinical data and lack of long term follow up. However, it provides insight into the endoscopic service for infantile hydrocephalus in Tanzania.

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