Childs Nerv Syst (2010) 26:937–943
DOI 10.1007/s00381-010-1130-1

ORIGINAL PAPER

Endoscopic third ventriculostomy in previously shunted children: a retrospective study

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Received: 11 November 2009 / Accepted: 14 March 2010 / Published online: 30 March 2010

Abstract
Purpose The aim of this study was to assess the mid-term results, success rates, and time-to-failure of secondary endoscopic third ventriculostomy (secondary ETV), as well as the complex management of preoperative and postoperative cares.
Methods To this purpose, a retrospective analysis of a pediatric population of 22 children who underwent endoscopic third ventriculostomy (ETV) after shunt malfunction (secondary ETV) was performed.
Results The failure rate, given by the percentage of new shunt replacement in the first 3 months after ETV, was 36%, with a mean time to failure of 14.3 days. All the failures were evident within 1 month after the ETV. Despite the small number of patients in our series, we found no significant correlation between ETV failure and both patient age and hydrocephalus etiology (p=0.47 and p= 0.78, respectively).
Conclusions In our experience, ETV secondary to shunt malfunction in pediatric patients has a success rate of 64%. As it is a safe and rapid treatment option even in emergency conditions, it is worth performing this procedure in previously shunted children.

Keywords Neuroendoscopy · Third ventriculostomy · Obstructive hydrocephalus · Ventriculoperitoneal shunt failure

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Introduction

The effectiveness of primary endoscopic third ventriculostomy (ETV) in obstructive hydrocephalus has proven to gain high success rates of shunt independency, with results ranging from 79% [1] to 87.1% [2].

ETV performed after shunt failure is termed "secondary ETV." Previous studies on pediatric [3–5] or mixed series [6–8] showed only slight differences in the rates of success between primary (74%) and secondary ETV (70%). While the success rates of the former depend on the etiology of the hydrocephalus, the latter is independent from the etiology. In 2002, Siomin reported an increase in the success of ETV for the treatment of post-hemorrhagic hydrocephalus from 60.9% to 100% in primary and secondary ETV, respectively [9]. In 2005, O'Brien showed that the success of ETV in post-hemorrhagic hydrocephalus varies from 27% in primary ETVs to 71% in secondary ETVs, while in post-infection hydrocephalus, those rates varied from 0% to 75% [8].

Further differences between primary and secondary ETVs are related to postoperative management. Indeed, little has been written on this topic. The aim of this study was to detect the success and failure rates of secondary ETV in our pediatric population and to describe a protocol for the management of those complex cases.

Materials and methods

The patients' data were collected and retrieved from our database of endoscopic procedures, containing collecting data from 1995 through 2008. In this time interval, 470 endoscopic procedures were performed, and 119 pediatric patients were retrieved from the pediatric population. In

these last 13 years, 22 children aged from 4 months to 14 years (mean age, 6.7 years) underwent endoscopic third ventriculostomy for shunt malfunction (Table 1). The etiology of hydrocephalus was malformative in five patients (two patients had complex multiloculated hydrocephalus and three had Dandy–Walker malformation), post-hemorrhagic in eight, tumor related in two, post-meningitic in three, and secondary to idiopathic aqueductal stenosis in four. All children had undergone a ventriculo-peritoneal (VP) shunt in early infancy. The selection of patients was based on specific characteristics evidenced at MRI T2 sagittal images: the patency and morphology of the prepontine cisterns, and above all, the anatomical features of the third ventricle floor, in order to be sure that there is enough space to make the hole. In the same period, in fact, we performed 58 shunt revisions for shunt malfunction in pediatric patients who did not present the characteristics mentioned above.

Shunt malfunction was diagnosed upon clinical and radiological data symptoms associated with images of ventricular dilatation on CT scan and with x-ray ventriculography for the detection of catheter integrity and patency (Fig. 1A). Most CT scans showed only slight differences in the ventricular dimensions compared to previous neuroradiological studies performed when the shunts were functioning, reflecting the high sensitivity and fragility of these patients.

Twenty patients had mechanical impairment of the shunt, and two had shunt infection with signs and symptoms of meningitis. The time between VP shunt placement and shunt failure ranged from 16 days to 14 years, with a mean time of 67.3 months (5.6 years). Seven out of 22 patients were scheduled for emergency surgery because of clinically manifested raised intracranial pressure signs.

Surgical treatment

Before scheduling ETV, temporary resolution of intracranial hypertension was achieved by an external ventricular drainage (EVD); this procedure gave us the time to perform MRI with cerebrospinal fluid (CSF) flow study to evaluate the patency of the ventricular cavities and the anatomy of the floor of the third ventricle for ETV feasibility assessment.

EVD was placed in 16 patients: a new ventricular catheter was placed in 10 patients, and in 4 out of those cases, an Ommaya reservoir was connected, too. EVD was placed from the shunt valve in 5 patients. In one patient, the external ventricular drainage was gained as the consequence of the exteriorization at the neck of the distal catheter of the previous shunt. In three patients, during the surgery for shunt removal, an Ommaya reservoir was placed without the positioning of an EVD, as a safe access in case of emergency. EVD was kept for a minimum of 3 days to a maximum of 15 days, with a mean time of maintenance of 4.7 days. In two cases, in fact, the preoperative CSF samples demonstrated an infection and the patients underwent intravenous antibiotic infusion for 12 and 15 days. In all the cases, we always used antibiotic impregnated ventricular catheters. No other antibiotic prophylaxis was used during EVD.

The removal of the previous shunt was performed at diagnosis in the two cases of shunt infection and in six cases with a catheter rupture and/or distal migration.

Ventricular dilatation for flexible endoscope navigation was achieved in patients with EVD by modulation of the height of the external drainage bag. MRI with CSF flow study was performed 3 days later to measure the ventricle's size, to assess the anterior protrusion of the third ventricle floor, and finally, to assess the patency of the aqueduct and fourth ventricle outlets (Fig. 1B).

In the six patients without EVD, MRI was performed in the first day of clinical observation.

ETV procedures were performed with a flexible 3.5-mm Storz endoscope in all patients. There were no intra-operative complications. After ventriculostomy, the shunt was consequently removed in ten patients. In four patients, the proximal catheter was left in place because it was hindered. Eighteen out of 22 patients had the complete removal of the previous shunt.

Postoperative management

Children were strictly monitored for clinical signs and symptoms of intracranial hypertension; a postoperative CSF-flow MRI was performed in order to assess the ventriculostomy patency and flow pulsatility. ICP monitoring was not recorded systematically because of the difficulties of the continuous monitoring in children, especially in the younger age groups. ICP monitoring was performed only in five patients. In all the other cases, ICP measurements were performed before lumbar taps and then recorded. Independently from the pathologic values of measured ICP (we considered alarm pressure at 20 mm Hg), decisions were conditioned almost exclusively from clinical signs and symptoms of raised intracranial pressure. Symptomatic patients were evaluated upon a basis of a point system scale, with a value of 1 point for each of the following five symptoms: headache, nausea and vomiting, irritability, lethargy, and bradycardia. The presence of at least three out of them and their clinical persistence for more than 48–72 h was considered the final point of clinical observation and the time of failure of ETV degree (Table 1).

During the observation time, ventricular taps were performed in 8 out of 22 patients, in particular, in three patients with reservoir without EVD, in four patients with EVD from the Ommaya reservoir, and in one patient with

Table 1 Secondary ETV in previously shunted children

	Age at ETV	Hydrocephalus etiology	Preoperative management	Post-ETV main symptoms	Post-ETV management	ETV revision	ETV failure time
1	8 years	Post-hemorrhagic	Exteriorization of distal catheter	Headache, vomiting, and bradycardia	Lumbar puncture		4 days
2	3 years	Post-hemorrhagic	EVD with Ommaya	Headache, irritability, and vomiting	Reservoir taps		12 days
3	2 years	Malformative	Reservoir positioning	Lethargy and vomiting	Reservoir taps		20 days
4	5 years	Post-hemorrhagic	EVD from valve	Headache, vomiting, lethargy, and bradycardia	Reservoir taps		2 days
5	3 years	Neoplastic	Reservoir positioning		Reservoir taps		
6	7 years	Post-meningitis	EVD with Ommaya		Reservoir taps		
7	8 years	Post-hemorrhagic	EVD with Ommaya		Reservoir taps		
8	4 months	Aqueductal stenosis	Reservoir positioning	Headache and vomiting	Lumbar puncture		19 days
9	11 years	Malformative			Lumbar puncture		
10	14 years	Aqueductal stenosis		Headache, irritability and lethargy	Lumbar puncture		28 days
11	6 years	Neoplastic	EVD	Lethargy, bradycardia	Lumbar puncture		10 days
12	4 years	Aqueductal stenosis	EVD		Lumbar puncture		
13	10 months	Malformative	EVD from valve		Lumbar puncture	ETV revision after 14 days	
14	12 y	Post-meningitis	EVD with Ommaya	Headache	Reservoir taps	ETV revision after 20 days	
15	5 y	Malformative	EVD		Lumbar puncture		
16	7 y	Aqueductal stenosis			Lumbar puncture		(ETV revision 3 years later)
17	3 years	Post-hemorrhagic	EVD from valve		Lumbar puncture		
18	6 years	Neoplastic	EVD		Lumbar puncture		
19	9 years	Malformative	EVD	Headache, vomiting, and bradycardia	Lumbar puncture	ETV revision after 15 days	20 days
20	11 months	Post-hemorrhagic	EVD from valve		Lumbar puncture		
21	3 years	Post-hemorrhagic	EVD from valve	Vomiting	Lumbar puncture	ETV revision after 7 days	
22	4 years	Post-meningitis	EVD		Lumbar puncture		

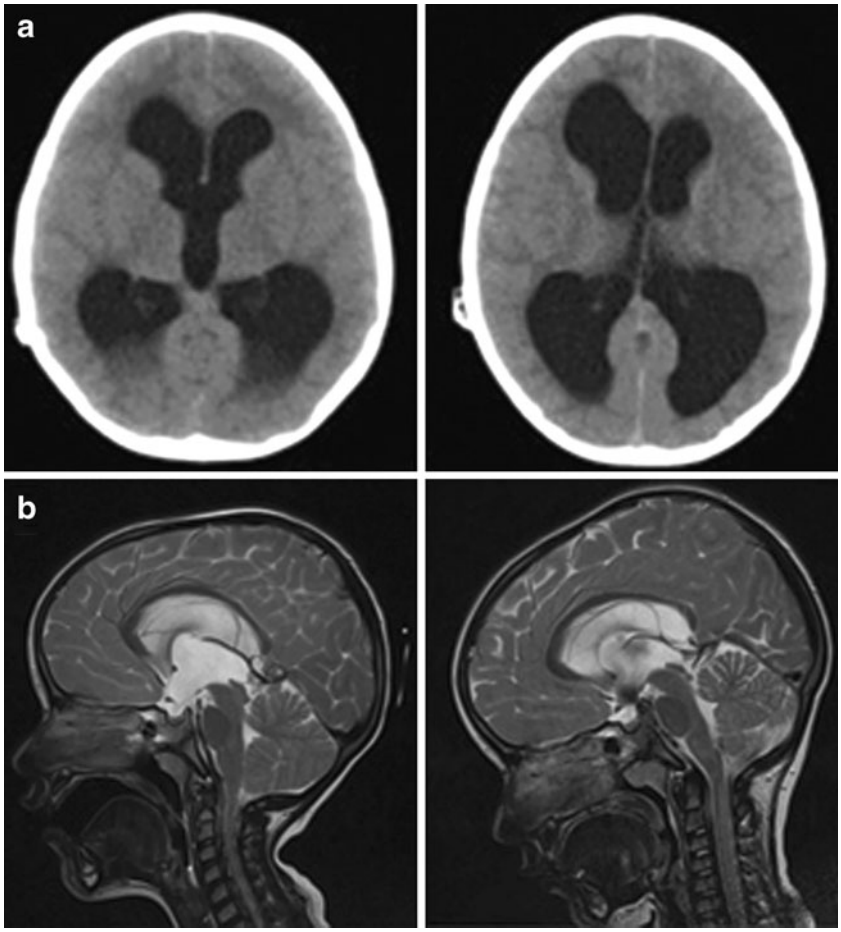

Fig. 1 **a** CT scan showing ventricular dilatation of both the lateral and the third ventricles. The ventricular catheter is not visible because it migrated out from the cranial hole in the subcutaneous space. **b** Postoperative T2-weighted sagittal MRI. Ventricular diameters are normal. On the right, a good CSF flux is perceivable

EVD from the valve of the previous shunt (Table 1). In the other 14 patients, only lumbar punctures were performed. It is worth noting the different management we kept during years. In the past, we used to perform ventricular taps in the management of post-ETV time. After some consecutive ETV failures (cases 2–4 and 8), we abandoned this maneuver because it was supposed to reduce the flow throughout the ventriculostomy. On the other hand, we performed more and more ventricular taps, by removing a volume of 30–40 ml of CSF/day in order to force the CSF flow at the ventriculostomy through the stoma site by enabling a gradual adaptation of the CSF absorption pathways.

In the early follow-up period, re-ventriculostomy was performed in three cases in which a partial obstruction of the stoma was detected on the early postoperative MRI (patients 14, 19, and 21).

One re-ETV was performed 3 years after the first procedure with good outcome.

The follow-up period of patients who received benefit from ETV after shunt failure had averaged to 45.5± 5.3 months (12–122 months).

Results

The failure rate, given by the percentage of new shunt placement in the first 3 months after ETV, was 36.37% (eight shunt replacements). The mean time of failure was 14.3 days, ranging from 2 to 28 days. Clinical symptoms, evaluated with the five-point score (Table 1), were the main decisional issues and were independently assessed from the stoma patency revealed at the CSF-flow MRI (Fig. 2).

MRI was useful for the assessment timing of a second endoscopic treatment performed with enlargement of the previous stoma in cases in which there was a weak radiological flow and no clinical relief.

There was no morbidity associated with the neuroendoscopic procedure and the follow-up after secondary ETV ranged from 12 to 122 months, with only one child needing a ventriculostomy revision after 3 years.

The correlation between ETV success rate, patient's age at surgery, and hydrocephalus etiology were tested with a χ^2 test, considering a $p<0.05$ statistically significant. Although a series of 22 patients is small and does not allow to achieve any broad conclusion, we could infer that

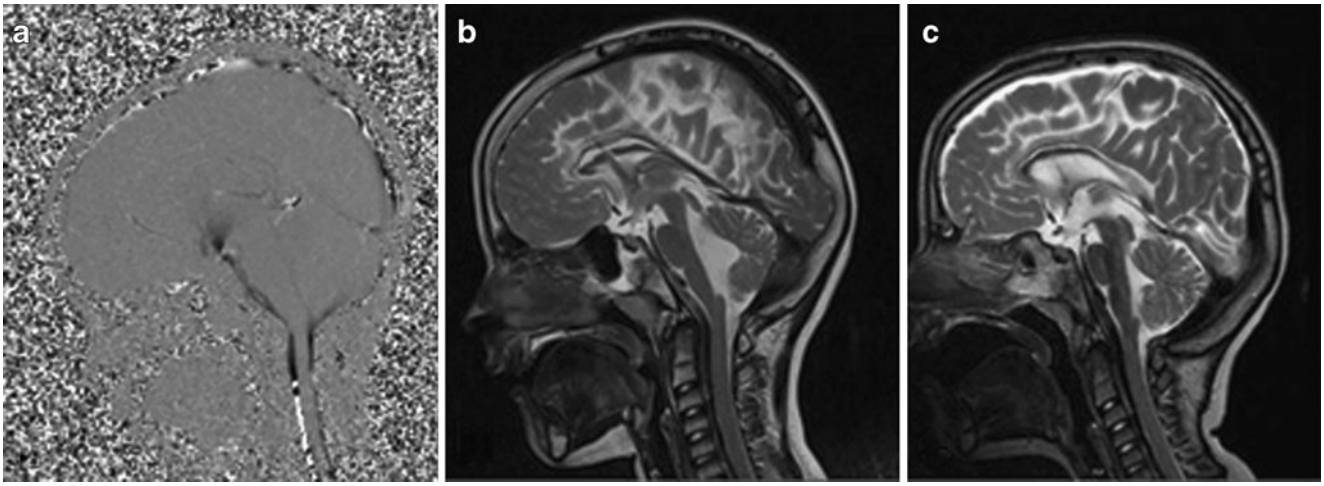

Fig. 2 This is the case of a child with clinical and radiological signs of hydrocephalus, although an apparently functioning ETV. **a** CSF-flow MRI confirming the patency of the previously performed ventriculostomy. **b** Preoperative T2-weighted sagittal MRI, with an evident dilatation of the fourth ventricle, obliteration of the prepontine and retrocerebellar cisterns, and displacement of the brainstem. **c** T2-weighted sagittal MRI after VP-shunt implant, showing the reduction of the fourth ventricle and the normalization of the brainstem and cerebellar structures

neither age (p=0.78) nor hydrocephalus etiology (p=0.47) statistically affect secondary ETV success.

On the basis of our experience, the use of neuroendoscopic procedures for shunt malfunction achieved shunt independence in 63.64% of cases.

Discussion

Primary ETV in obstructive hydrocephalus has proven to be effective, with an overall reported rate of shunt independency ranging from 79% [1] to 87.1% [2].

As postoperative failure may occur generally early, a strict follow-up especially in the first year after surgery is mandatory. The follow-up should be maintained anyway in these patients, as delayed failures have been described, causing acute malfunction of the ETV, leading even to sudden death [7, 10].

Particular considerations pertaining to ETV and hydrocephalus in children must be discussed, especially in the first 2 years of life.

Data from literature suggest that the clinical response to ETV of adult patients with obstructive hydrocephalus is different from that of children, and differences are secondary to the hydrocephalus age, CSF dynamics, and changes in brain viscoelastic properties [1]. The success rate of ETV in the pediatric population was reported with percentages varying from 46% [11] to 70% [3] at the first treatment, underlying a clear negative impact of age on ETV failure rate that gradually increases in the first months of life [3]. ETV was considered as the treatment of choice in obstructive hydrocephalus, even before the first 2 years of life, with a success rate ranging from 42% to 43% [12, 13] to 57% [14]. A reduction of shunted population up to 21% is reported [15] as a consequence of ETV as primary treatment in infants with hydrocephalus. In both adults and children, ETV is a safe and effective procedure, and early infancy should no longer be considered as a contraindication [8]. Also repeated, ETV has been shown to be effective in primary ETV procedures [16]. Recent reports tried to define statistical models to predict the probability of ETV success on the basis of a child's individual hydrocephalus features [17]. On the other hand, Takahashi [18] in 2006 reported in a long-term follow-up that the cerebral development of infants with inadequate cortex development is better in the group of patients treated with shunt instead of ETV.

The risk of VP shunt malfunction is well known, ranging from 25% to 40% in the first year after shunt placement and remaining at 4% to 5% per year [19]. Shunt failure is almost unavoidable during a patient's life, with 81% of shunts requiring revision after 12 years [20]. Shunt infection occurs at a relatively high frequency, with typical reported rates of 5% to 10% [21] but with values rising up to 19% in a small series [22]. Although ETV does not lack infective complications, the reported infection rates are approximately 2%. ETV avoids shunt complications such as mechanical malfunction, abdominal adhesions with consequent impairment of the absorptive properties of the peritoneum, slit-ventricle syndrome, and CSF overdrainage [23].

Reliable long-term data about the effectiveness of ETV are not as readily available as for VP shunt implants. Cinalli reported that 72% of the stereotactically performed ETVs

for triventricular hydrocephalus are still functioning after 6 years [3]. Takahashi reported a follow-up of up to 6 years in children treated with ETV under 9 months old: in the group of patients with abnormal cortex development, ETV failed and a VP shunt was placed [18]. ETV alone was not sufficient to improve inadequate cerebrum development, and early shunting resulted more useful for achieving cerebral development recovery. Further analyses with a long-term follow-up are required.

Several authors consider ETV a successful treatment in the management of shunt malfunction or infection in patients with obstructive hydrocephalus. Studies on pediatric [3–5, 24] or mixed series [6–8] showed only slight differences in the rates of success between primary (74%) and secondary (70%) ETV. But if we highlight hydrocephalus etiology, results are more challenging to be interpreted. While the success of the former depends on the etiology of hydrocephalus, the latter seems to be independent. Siomin in 2002 reported an increase in ETV efficacy in post-hemorrhagic hydrocephalus from 60.9% to 100% in primary and secondary ETV, respectively [9]. In 2005, O'Brien reported that the success of ETV in post-hemorrhagic hydrocephalus varies from 27% in primary ETVs to 71% in secondary ETVs, while in post-meningitic hydrocephalus, the success of primary and secondary ETVs varies from 0% to 75%, respectively [8].

These results are probably related to the fact that post-hemorrhagic or post-meningitic hydrocephalus at its beginning is communicating due to obstruction of the subarachnoid spaces and arachnoid granulations with infective/hemorrhagic debris. The procedure also may induce an acquired aqueductal stenosis through continuous CSF diversion, increasing the likelihood of success of an ETV after a shunt malfunction. Similarly, the failure of the primary ETV in infective/hemorrhagic cases is explained by the blockade of the CSF subarachnoid spaces and arachnoid granulations by debris and membranes, which counteract the mechanism of the ETV.

In our experience, we treated 22 children from 4 months to 14 years of age. All failures in our group were clinically detectable within 1 month after the ETV and were managed with a new VP shunt. Three cases required a second ETV procedure in the first month.

Analyzing the impact of age on secondary ETV success and considering an homogeneous pediatric population, no correlation between age and ETV success seems detectable, with the success rate homogeneously spread in all pediatric ages, even when we excluded etiological factors [8].

Failure of the secondary ETV does not seem associated with hydrocephalus etiology or the patient's age. Failure of secondary ETV seems rather to be linked to the blockage of the CSF subarachnoid spaces and arachnoid granulations by debris and membranes, which continues to be the major obstacle to CSF flow, similar to the mechanism responsible of the failure of the primary ETV. In our experience, we also found that the practice involved in the first years of performing these endoscopic procedures and having minor experience with ETV in pediatric patients contributed to ETV failures. In the following years, the practice of creating a large ventriculostomy or enlarging the small ones, the systematic spinal lumbar taps to control symptoms in the first days, and the experience acquired gave better clinical results.

We also should address some issues on the heterogeneity found in literature about postoperative management after a secondary ETV. ICP monitoring, the use of an external ventricular drainage, the amount of cerebrospinal fluid that needs to be taken, the need for CSF spinal taps, and most importantly, the parameters used to declare that the ETV has failed, leading to the insertion of a new shunt, are not as clear and shared.

Jones suggests CSF removal with spinal taps or reservoir taps according to raised intracranial pressure, implying a continuous bedside monitoring of intracranial pressure [5]. Boschert recommends an external ventricular drainage to be kept in place for several days and opened only for emergency clinical signs [6]. Cinalli measured ICP in the early postoperative days and investigated the role of lumbar puncture to allow a faster normalization of the ICP, suggesting a cycle of one to three lumbar punctures being performed before assuming that ETV failed in patients who remain symptomatic and show ventricular dilation after ETV [25].

In our series, fourteen patients with secondary ETV underwent spinal taps for a mean period of 2.42 days. In eight cases of ETV failure, spinal or ventricular taps were obtained for a mean time of 12.4 days in an attempt to improve the CSF flow through the ventriculostomy. In two cases, no taps were deemed necessary because of the complete remission of the clinical signs and symptoms of raised ICP.

In our experience, we agree that the rationale of the post-ETV management is the progressive reduction of the artificial removal of CSF in order to gently force the flow through the stoma. The optimal reduction should be done in 3 days. If the patient needs taps for more than 3–4 days, the ventriculostomy probably will never work.

Conclusions

We are aware that the limitations of our study lie in its observational retrospective nature and sample's size. However, ETV seems to be a safe and minimally invasive procedure with few complications and high success rates in pediatric patients with shunt malfunction. Compared to

primary ETV, secondary ETV does not seem to be correlated with age or the etiology of hydrocephalus.

ETV secondary to shunt malfunction in pediatric patients has, in our experience, a success rate of 63.64% in the first month after the procedure. As it is a safe and rapid procedure even in emergency conditions, it is worth to be attempted in previously shunted children in order to offer them the best chance of shunt independence. Most failures are detectable early, but long-term follow-up is mandatory.

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