

Result of Endoscopic Third Ventriculostomy for Hydrocephalus Caused by Cerebral Aqueductal Stenosis in Infant Patients

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Abstract

Background Endoscopic third ventriculostomy (ETV) is one of the important management options for hydrocephalus, especially obstructive hydrocephalus. The result of ETV in different age and etiology is quite variable according to the different authors. The study was carried out to assess the success rate of ETV as a treatment for this type of hydrocephalus in infant age group.

Material and Methods The study was done prospectively on infants (age \leq 12 months) admitted in the period from January 2009 to June 2013 suffering from obstructive hydrocephalus due to cerebral aqueductal stenosis (CAS). After making the diagnosis, we counseled the patient party about the probability of high failure rate of ETV in this age group and probable necessity of a second operation. With the informed consent, we performed ETV in these cases and followed up regularly in postoperative period. Control neuroimaging studies were done whenever needed (suspected failure of ETV and suspected stomal block).

Results Total 17 infants were studied. Average follow-up was 18.7 months. Twelve infants were between the age group of six and under six months while five were above six months to one year old. Fourteen (82.35%) out of 17 patients showed overall clinical improvement. Clinical improvement was seen in two (66.6%) infants aged 2 months or younger, three (75%) aged 2⁺ to 3 months, five (100%) aged 3⁺ to 6 months, and four (80%) aged 6⁺ months to 1 year. Two patients had bleeding during surgery. Three developed CSF leak through the burr hole. In one case (the patient's age was 2 months), the stoma was found blocked, and hydrocephalus returned 9 weeks after ETV. In two patients, in whom VP shunt was needed, "failed ETV" was obvious in early postoperative period. In this series, average ETV success score (ETVSS) was 52.35 (range: 40–70) and overall success rate was 82.35%. This indicates that ETVSS does not correlate with the outcome of ETV in infants with CAS.

Conclusion Outcome of ETV for hydrocephalus from CAS in infant is quite good and ETVSS does not correlate with the outcome.

Keywords

- ▶ endoscopic third ventriculostomy
- ▶ obstructive hydrocephalus
- ▶ cerebral aqueductal stenosis
- ▶ ETV success score
- ▶ results

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Introduction

The most common therapeutic options used to treat hydrocephalus (HCP) include ventriculoperitoneal (VP) drainage and neuroendoscopic surgery. Although there are pros and cons for each option, VP shunt techniques have high complication rate (20–80%).^{1–3} Endoscopic third ventriculostomy (ETV) is well accepted for obstructive hydrocephalus of various etiologies.^{4–10} It is considered as a better alternative to shunt surgery in obstructive HCP. Though some authors advocate ETV in patients of all ages,^{1–3,11} others showed that patients younger than 1 year have a higher failure rate for ETVs compared with older children.¹² Therefore, we present our experience of ETV in obstructive HCP caused by congenital cerebral aqueductal stenosis (CAS), comparing between the patients below six months and above.

Material and Methods

The present study was done prospectively in the Neurosurgery Department, Dhaka Medical College Hospital, Islami Bank Central Hospital, and Ibn Sina Hospital, Dhaka, Bangladesh, on infants (age \leq 12 months) with obstructive HCP due to CAS, admitted in the period from January 2009 to June 2013. Failed VP shunts in CAS cases were also included. CAS patients associated with arachnoid cyst, meningocele, or myelomeningocele were excluded (to make the study homogeneous) where CAS caused triventriculomegaly. The patients who were not available for at least 6-month follow-up were not included in the study. (Regular follow-up in a developing country such as Bangladesh is a challenging job. Availability of mobile and video phone calls helped in regular follow-ups in these cases. Sometimes we had to pay some money to the parents for meeting up their traveling cost.) For diagnosis of HCP, ultrasonogram (USG), computed tomography (CT) scan, and magnetic resonance imaging (MRI) of the brain were used. USG of the brain was done in eight cases as a screening test by the attending physician. CT scan of the brain was done in 15 cases and MRI was done only in 2 cases because the parents were unwilling to let the infants undergo general anesthesia. After making the diagnosis, we counseled the patient's parents about the probability of high failure rate of ETV in this age group and probable necessity of a second operation. With the informed consent, we performed ETV in these cases. The study was carried out to assess the success rate of ETV as a treatment for this type of HCP in such age group. Patients were followed up postoperatively from days 1 through 7; then weekly assessment for 2 months, and then monthly assessment for 12 months followed by 3 monthly assessment of the head circumference, anterior fontanelle, cerebrospinal fluid (CSF) leak, wound infection, fever, or any other complaints. ETV was considered as successful if the patient improved clinically. Control neuroimaging studies were done whenever needed (suspected failure of ETV and suspected stomal block) or after 6 months to 1 year.

Operative Technical Points

The operative technique of ETV is standardized and is not mentioned here. After making an opening in the third ventricular floor, endoscope was advanced below through the opening to inspect further. If an imperforate membrane of Lilliequist was identified, lying beneath the floor of the third ventricle was opened under direct endoscopic visualization.

If any hemorrhage was encountered during the procedure, copious warm fluid irrigation was used until all bleeding was visibly stopped and the ventricular CSF was clear. We used careful intermittent closure of outflow channel to create tamponade effect. In one patient, in whom irrigation failed to clear vision, blood-stained CSF and normal saline were aspirated by a Fogarty catheter whose tip was cut off before aspiration. After removal of endoscope with sheath, wound was closed accordingly. First dural closer was done, and then bone dust and spongostan were placed at the burr hole. Pericranial-aponeurotic layer was closed carefully. Finally skin was closed with interrupted stitches.

Results

Total 17 infants were studied, out of these 8 (47%) were male and 9 (53%) female. Follow-up range was 7 to 37 months (average: 18.7 months). Details of all patients are shown in **►Tables 1 and 2**. There were seven (41%) infants aged 3 months or younger and three (17.6%) were 2 months old or younger. Twelve (70.6%) were 6 months or younger whereas five (29.4%) were older than 6 months to 1 year. No one was born prematurely. There was history of febrile illness in the mother during pregnancy among three infants. Ten (58.8%) infants were the first issue of the parent. One patient had history of mild birth asphyxia. Two infants had completely healthy twin siblings. Mild to moderate underweight was present in six (35%) cases. Gross protein energy malnutrition (marasmus) was present in one case for which concurrent nutritional therapy had to be given during the perioperative period of ETV. Low-birth weight (LBW) was present in three (17.6%) cases. Three, four, five, and five infants were aged 2 months or younger, 02⁺ to 03 months, 03⁺ to 06 months, and 06⁺ months to 1 year, respectively. There was no history or event of pre-ETV intraventricular hemorrhage or infection (even in failed VP shunt cases). Fourteen (82.35%) out of 17 patients showed overall clinical improvement (**►Figs. 1–3**). Clinical improvement was seen in two (66.6%) infants aged 2 months or younger, three (75%) aged 2⁺ to 3 months, five (100%) aged 3⁺ to 6 months, and four (80%) aged 6⁺ months to 1 year. Out of three LBW infants (all aged < 2 months), one (33.3%) patient failed ETV. Average age of this series was 5.5 months. The mean age of the patients with success was 5.6 months. Average ETV success score (ETVSS) of the series was 52.35. Average ETVSS was 47.5 (range: 40–50) and 64 (60–70) in infants aged 6 months or younger and 06⁺ to 12 months age group, respectively.

Two patients had bleeding during surgery. In one case, continuous irrigation along with endoscopic tip pressure and pressure by inflated Fogarty balloon were needed to

Table 1 Details of patients: 0–6 months of age

Age (mo), sex and, figure	Diagnosis/ETVSS	Pre-ETV VP shunt	H/O birth asphyxia, premature delivery, LBW, PEM	Perioperative complication	Postoperative complication	F/U (mo)	ETV failed/Stoma block	Reoperation
4/F (► Fig. 1)	CAS/50	–	–	–	–	14	–	–
4/F	CAS/50	–	–	Bleeding	CSF leak–LP failed	32	Failed ETV	Re-ETV failed: VP shunt done
3/F	CAS/50	–	–	–	–	27	–	–
23 d/F	CAS/40	–	LBW	–	–	15	–	–
2/F	CAS/50	–	–	–	–	10	–	–
5/M	CAS/50	–	–	–	–	7	–	–
5/M	CAS/40	Failed VP shunt	–	–	–	23	–	–
3/F (► Fig. 2)	CAS/50	–	–	Bleeding	CSF leak–healed by LP	37	–	–
3/F	CAS/50	–	–	–	Fever	09	–	–
3/M (► Fig. 3)	CAS/50	–	–	–	–	11	–	–
5/F	CAS/40	Failed VP shunt	–	–	–	12	–	–
2/M	CAS/50	–	LBW	–	–	21	Stoma blocked-9 wk after ETV	Re-ETV failed: VP shunt done

Abbreviations: CAS, cerebral aqueductal stenosis; CSF, cerebrospinal fluid; ETV, endoscopic third ventriculostomy; ETVSS, ETV success score; F/U, follow-up; F, female; LBW, low-birth weight; LP, lumbar puncture; M, male; PEM, protein energy malnutrition; VP, ventriculoperitoneal.

stop bleeding. Force aspiration through sheath channel and “cut tip Fogarty catheter” needed to make vision clear in other case. No procedure had to be abandoned. Three patients developed CSF leak through the burr hole. Leak stopped spontaneously in 3 days in one patient. One patient needed three times lumbar puncture whereas in the remaining patients (there was perioperative hemorrhage), repeated lumbar puncture and re-ETV failed to resolve the problem and finally VP shunt was needed. In one patient (age 2 months), the stoma was found blocked and HCP returned 9 weeks after ETV. As re-ETV failed in this case, the authors went for VP shunt that resolved the problem. In another case in which ETV failed (ETV was done at the age of 9 months for complication of previously done VP shunt), we had to return to VP shunt. Where VP shunt was needed, “failed ETV” was obvious in early postoperative period cases in two cases, and in the remaining cases, features of HCP reappeared 9 weeks after ETV due to stomal block. The patients’ ages were 2, 4, and 9 months at the time of ETV when VP shunts were needed. We faced no mortality, diabetes insipidus (DI), or endocrine abnormalities. Transient postoperative fever was found in three cases. ETV was done in five cases in which previously done VP shunt was failed; four out of five responded well with ETV and one needed repeat VP shunt.

Discussion

New interest developed in the use of ETV for the treatment of obstructive HCP along with advanced fiberoptic and lens technology. In recent time, the better success of third ventriculostomy could be attributed to better patient selection and improvements in endoscope, better imaging, advanced surgical technique, and instruments. ETV is considered treatment of choice in obstructive HCP. It is also now used in some communicating HCP, such as normal pressure HCP by some authors. Although ETV can be used in selected patients of all types of HCP, the success of ETV in obstructive HCP is better than that in communicating HCP.⁴

Obstructive HCP occurs commonly due to congenital CAS, posterior third ventricle tumor, cerebellopontine angle tumor, and other posterior fossa tumor of obstructive variety. HCPs following cerebellar infarct, Dandy-Walker malformation, syringomyelia with or without Chiari’s malformation, shunt malfunction, encephalocele, craniosynostosis, and intraventricular hematoma are usually of obstructive type. Myelomeningocele with HCP, slit ventricle syndrome, and multiloculated HCPs are also of obstructive type. HCP secondary to intraventricular hemorrhage, ventriculitis, meningitis, and postoperative cases after complete excision of mass lesions are of communicating variety. Normal pressure HCP is of communicating type. When

Table 2 Details of patients: 6+–12 months of age

Age (mo) and sex	Diagnosis/ETVSS	Pre-ETV VP shunt	H/O birth asphyxia, premature delivery, LBW, PEM	Perioperative complication	Postoperative complication	F/U (mo)	Stoma block	Reoperation
9/M	CAS/60	VP shunt block with downward migration	PEM (marasmus)	–	–	35	–	–
12/M	CAS/70	–	–	–	CSF leak: stopped spontaneously	21	–	–
9/M	CAS/60	CSF leak at abdominal end of VP shunt (at the age of 3 mo)	Mild birth asphyxia	None	None	15	ETV failed (failed to improve symptoms)	Re-VP shunt on the opposite side
12/F	CAS/70	–	–	–	–	36	–	–
9/M	CAS/60	VP shunt: at the age of 2 mo	–	–	Fever	13	–	–

Abbreviations: CAS, cerebral aqueductal stenosis; CSF, cerebrospinal fluid; ETV, endoscopic third ventriculostomy; ETVSS, ETV success score; F, female; F/U, follow-up; M, male; mo, month; PEM, protein energy malnutrition; VP, ventriculoperitoneal.

both the element of obstruction and the defect in the CSF absorption persist, it is complex HCP.^{4,13}

CAS is a common cause of obstructive (noncommunicating) HCP. It could be due to congenital stenosis, acquired idiopathic, postinfectious or posthemorrhagic, and stenosis secondary to tumor. In congenital stenosis, aqueduct could be narrow or

completely obstructed. Complete or near-complete obstruction usually presents in an early age, whereas if obstruction is partial, patients may be asymptomatic or present at a later age. ETV instead of a shunt placement is considered a better option for noncommunicating HCP secondary to congenital aqueductal stenosis. Results of ETV are better in congenital HCP

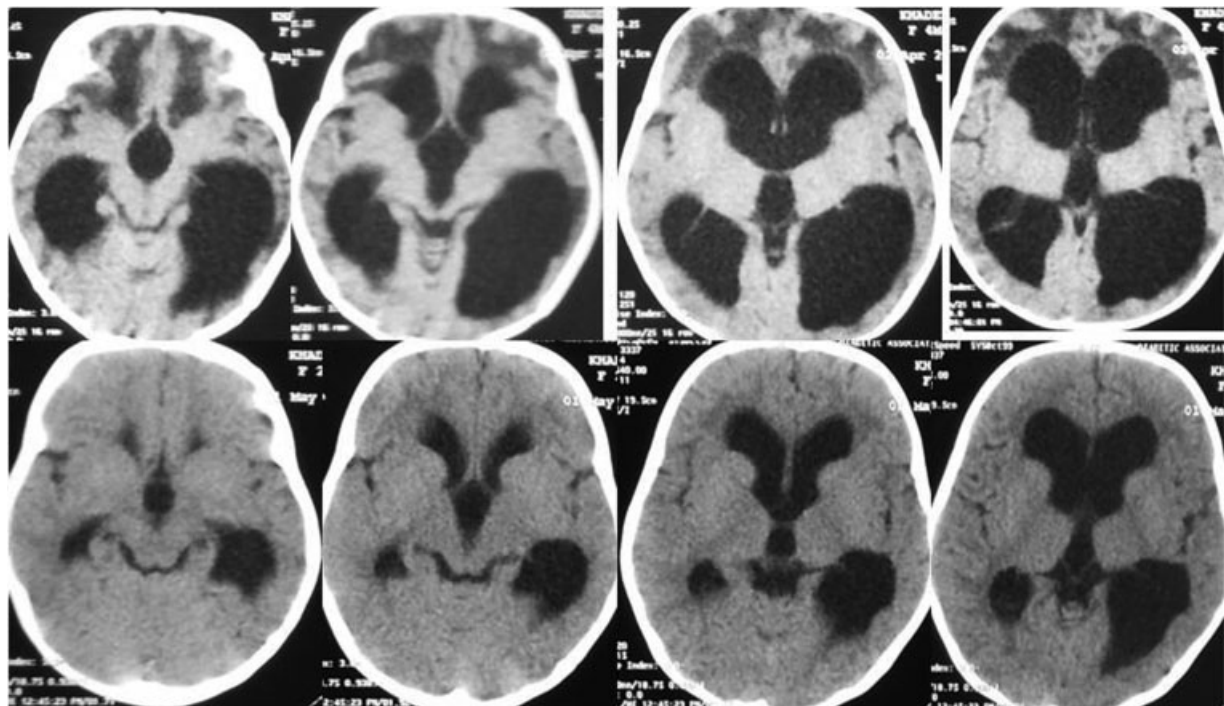


Fig. 1 Upper row—preoperative serial axial CT scan of brain showing triventriculomegaly at the age of 4 months. Lower row—postoperative serial axial CT scan of brain at the age of 2 years. CT, computed tomography.



Fig. 2 (A) Preoperative picture of patient with hydrocephalus. (B) Preoperative MRI sagittal section in T1W image showing triventriculomegaly with CAS. (C) Early postoperative picture of patient after ETV. (D) Late postoperative picture of patient with successful ETV. CAS, cerebral aqueductal stenosis; ETV, endoscopic third ventriculostomy.

due to aqueductal stenosis as compared with posthemorrhagic or postinfective HCP.^{4,5} Perioperative neural injury, such as thalamic, fornix, hypothalamic, and midbrain, are not uncommon. Intraoperative bradycardia and hemorrhages including fatal hemorrhage due to basilar artery rupture are also reported. Attempts to perforate the ventricular floor can lead to bleeding, especially in HCP following an infection and hemorrhage. From our experience, all can be easily avoided by appropriate placing of burr hole and careful surgical manipulation of endoinstruments and neurostructures. Significant side movement should be avoided to prevent bleeding due to an injury to structures, such as the fornix and veins at foramen of Monro. We did not face any neural damage except controllable bleeding per operatively. Rarely, blood might trickle from burr-hole site into the ventricle; proper hemostasis must be achieved before entering the ventricle.⁴ Perioperative bleeding is seen in about 3 to 8%^{3,14} of cases. Aspiration of clots and thorough irrigation of ventricular cavity should be done after bleeding stops; these measures can reduce chances of stoma closure.³ Bradycardia may occur from raised ICP due to inappropriate irrigation and brainstem stretching.⁴

Central nervous system infections, fever, stoma block, CSF leak, and postoperative intracranial hematomas were also seen. Postoperative mortality is also reported.^{15,16} DI, hemiparesis, gaze palsy, memory disorders, altered consciousness, weight



Fig. 3 (A) Preoperative picture of patient with hydrocephalus. (B) Pre-ETV CT scan axial sections showing triventriculomegaly due to CAS. (C, D) Early postoperative pictures of patient with successful ETV. CAS, cerebral aqueductal stenosis; CT, computed tomography; ETV, endoscopic third ventriculostomy.

gain, precocious puberty, and abnormal prolactin levels can occur after ETV. However, abnormal prolactin levels are not clinically significant.¹⁷ DI was reported by Choi et al¹⁸ in 3% cases. Chronic subdural hematoma or subdural hygroma can occur after ETV. Rare complications such as postoperative hyperkalemia, severe parkinsonism, acute respiratory alkalosis, and tachypnea can occur after ETV. Overall complication rate after ETV is about 2 to 15%, but the permanent complications are few.^{4,19-21}

Postoperative fever could occur due to use of electrocautery, which should be avoided. We faced postoperative fever in three cases that subsided spontaneously. Postoperative CSF leak could be avoided by plugging cortical and dural opening by Gelfoam, direct dural closure, especially in large ventriculomegaly in infants, or by using artificial dural substitute and tissue sealant in at-risk patients. Postoperative CSF leak can also be reduced by galeal-pericranial flap.^{4,22} Incidence of CSF leak after ETV have varied between 2 and 7%. In the series of Yadav et al, CSF leak occurred in 16% patients in whom half of the cases of CSF leak stopped spontaneously. The causes of persistent leak include closure of stoma, failure of closure of the dura, thinned-out scalp, and thin cortical mantle in patients with gross HCP. Dura closer should be done properly, especially in patients with gross HCP with thin cortical mantle.³ Postoperative complications in the ETV patients usually occur within 1 month after operation. However, delayed complications including stoma block can occur. Delayed stoma block, though very rare, can be fatal.²³ Intraoperative observation of thickened arachnoid membranes at the level of the interpeduncular cisterns at the time of ETV should be considered a significant risk of stoma block.²⁴ Postoperative failures usually occur early; regular clinical and radiologic follow-up must be performed, especially in the first years after the ETV.²⁵ According to Mohanty et al,²⁶ reclosure of the stoma because of gliosis and scarring has been observed in 6 to 15% of ETV failures. The high rate of reclosure in infants can be explained as follows: as CSF absorption is impeded, there is a greater tendency for the development of new arachnoid membranes in infants, and there is also growth of gliotic, ependymal, and scar tissue.¹² ICP monitoring with or without external ventricular drainage during the immediate postoperative period after ETV could be required in patients who continue to have clinical features of raised ICP or failed to show an improvement after ETV. Few patients fail to show an improvement despite patent stoma after ETV. CSF drainage by lumbar puncture helps by increasing the compliance and the buffering capacities of the spinal subarachnoid spaces. It probably decreases the CSF outflow resistance from the ventricular system, facilitates the decrease in the ventricular volume, and allows faster permeation of CSF in the intracranial subarachnoid spaces. A cycle of one to three lumbar punctures should always be performed in patients who remain symptomatic after ETV, before ETV is assumed to have failed.^{4,27-29}

A reduction in the ventricular size detected soon after ETV is associated with a good clinical outcome. This decrease in ventricular size continues during the first few months after surgery. Reduction in the size of third ventricle width is more than that in lateral ventricle size width after successful

ETV. Magnetic resonance ventriculography was effective in assessing subarachnoid space and stoma patency after ETV. T2-weighted turbo inversion-recovery magnetic resonance images can detect flow-void sign better. Cine phase-contrast (PC) MRI is useful even if no flow void is seen. Cine PC MRI may be used to determine the patency of the stoma and may be used in follow-up.^{4,24,30,31}

Determining the best candidates for ETV has been difficult, with conflicting reports on who are the best candidates, particularly regarding the effect of age and etiology. Reports have indicated that outcome is a function of age^{23,32,33} independent of age,^{1,34} a function of etiology,^{1,35-37} or a function of both age and etiology.³⁸ More recent evidence from larger, and in one case, multicenter series has supported the finding that age is the main determinant of outcome with younger children, particularly neonates, faring worse.^{39,40} The other standard technique of CSF diversion, a CSF shunt, has rarely been compared with ETV, with results suggesting no difference⁴¹ or a slight advantage in terms of cost-effectiveness for ETV.⁴² CSF shunt outcomes are also known to be influenced by age, with younger children also faring more poorly.⁴³⁻⁴⁵

There are controversies regarding the success of ETV in infants. Some authors reported poor results, especially in neonates and in infants younger than 2 months.⁴ Shim et al⁴⁶ suggested that simultaneous ETV and VP shunt should be performed in infantile HCP due to poor results of ETV alone. The higher clinical success rate of ETV in the series by Yadav et al was 83.7%.³ Success rates of 71%,⁴⁷ 64%,⁴⁸ and 85%⁴⁹ were observed in other studies done in infants whereas the success rates varied from 76¹⁴ to 91.5% in other age group patients.¹⁸ They found that failure rate of ETV in LBW premature infants was higher (60%) as compared to full-term normal-birth-weight infants (12.3%).³ In the case of children under 1 year, long-term shunt independence was 75% in the series by Stan et al.⁵⁰ The success of these patients was influenced much by the technique used. In the absence of plexus coagulation, the success rate of the operation is significantly reduced. The study by Warf et al proved the difference in the success rate with 48.6% in patients in whom they practiced only ETV compared to 81.9% in those in whom they practiced ETV and plexus coagulation both.⁵¹ Kulkarni et al⁵² reported the relative higher risk of initial failure in ETV than shunt in children. The relative risk becomes progressively lower for ETV after about 3 months. Patients could experience a long-term treatment survival advantage after an early high-risk period of ETV failure as compared to shunt. They observed that it might take several years, however, to realize this benefit. There have been several studies of the effectiveness of ETV in children under two years of age. Kadrian et al reported a strong effect of the patient's age on outcome.⁴⁰ The authors reported the percentages of patients "presumed to have a functioning ETV after 5 years" as follows: 41% in patients 1- to 6-month old at the time of surgery, 58% in patients 6- to 24-month old, and more than 70% in patients older than 24 months. These results correspond to the data reported by other authors.³² However, Javadpour et al⁴⁷

Table 3 Comparative success rate of ETV in aqueductal obstruction by some published series

Series	No. of patients	Age group	Result of success rate (%)
Fritsch et al (2005) ¹	3	Pediatric (< 1 y)	100
Cinalli et al (1999) ¹¹	119	Pediatric	72
Zohdi et al (2013) ¹²	08	Pediatric	12.5
Gangemi et al (2007) ²⁵	88	Pediatric plus adult	87
Drake (2007) ³⁹	107	Pediatric	65 and 52 success rates at 1 and 5 y, respectively
Tuli et al (1999) ⁴¹	32	Pediatric	56
Goarayeb et al (2004) ⁴⁸	11	Pediatric (< 1 y)	55
Warf et al (2005) ⁵¹	153	Pediatric	53 in all patients and 70 in aqueductal obstruction
Fukuhara et al (2000) ⁵⁷	37	Pediatric plus adult	68
Bognar et al (2005) ⁵⁸	76	Pediatric plus adult	71
Koch-Wiewrodt and Wagner (2006) ⁵⁹	13	Pediatric (< 1 y)	54
Sacko et al (2010) ⁶⁰	350	Pediatric plus adult	61

Abbreviation: ETV, endoscopic third ventriculostomy; y, year.

reported ETV success rate of 33% (continued patency during follow-up in 7 of 21 patients) and found that success depended on etiology rather than on patient age. Baldauf et al³⁸ reported 50% success rate in idiopathic aqueductal stenosis. Analysis of the success and failure of ETV in infants in the Netherlands confirmed that ETV should be considered as an initial treatment that carries a low risk of morbidity in these infants. As the immune system rapidly matures, postponing shunt implantation for several months or even weeks would make ETV worthwhile. Moreover, the authors noted that a second ETV should always be considered before shunt placement in young patients with a failed ETV, as the probability of ETV success rapidly increases 4 months after birth.⁵³ The largest study of ETV success in very young children was conducted in Uganda and it involved 153 children younger than 1 year.⁵⁴ The ETV success rate among these patients was 53%. The surgery success rates for patients with aqueductal obstruction were 70%. Fritsch et al,¹ reporting a 39% ETV success rate, presented ETV as an effective alternative for the treatment of obstructive HCP in infants younger than 1 year. Korean authors have reported simultaneous implantation of a VP shunt and ETV as the first choice of treatment for hydrocephalic patients younger than 1 year (83.9% success rate). Perhaps placement of the ventricular catheter in the prepontine cistern under endoscopic guidance reduces the risk of stoma closure and development of new arachnoid membranes.⁵⁵ On the other hand, these combined procedures do not provide patients with shunt independence and or freedom from shunt complications.¹² In the small series of ETV under the age of 6 months in obstructive HCP due to CAS, Zodi et al found very low success rate (only 12.5% cases were successful).¹²

Success rates of ETV in different series are shown in (► **Table 3**).^{1,11,12,25,39,41,48,51,56-60} ETSS is used to predict

the outcome of ETV before operation.⁵² In this series, average ETSS was 52.35 (range: 40–70) and overall success rate was 82.35%. This indicates that ETSS does not correlate with the outcome of ETV in infants with CAS. In our series, we did not find any impact of age on the success of ETV in CAS. In this small series, success rate of ETV in CAS under the age of 1 year was not low (82.35%) without choroid plexus coagulation. It is not well understood; result of success in ETV in obstructive HCP in infant was so variable in different series. During operation the authors frequently noticed extra-arachnoidal membrane after fenestration of third ventricular floor; the authors went for fenestration of that membrane(s) almost routinely to the anterior surface of the basilar artery. In future, larger series with more number of cases will probably answer the question preciously.

Conclusion

Outcome of ETV for HCP from CAS in infant is quite good with low risk of complication even in very young infant, and ETSS does not correlate with the outcome.

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