Obliteration of the choroid plexus after endoscopic coagulation

Clinical article

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Object. Endoscopic choroid plexus coagulation (CPC) with or without endoscopic third ventriculostomy (ETV) has been shown to be effective for selected patients with hydrocephalus. However, whether the effect of the coagulation is temporary and the choroid plexus regenerates or can be obliterated has remained largely unknown. The authors evaluate the effectiveness of CPC and report 3 cases of obliteration demonstrated by direct endoscopic observation.

Methods. The authors retrospectively analyzed the surgical results of patients with hydrocephalus primarily treated by CPC with or without ETV. Charts were reviewed for demographic data, clinical presentations, surgical therapies, and clinical outcomes.

Results. Eighteen patients with hydrocephalus were surgically treated using endoscopic CPC between July 2002 and July 2012. In 12 patients, ETV was concurrently performed. The etiology of hydrocephalus was posthemorrhagic in 5 patients, myelomeningocele in 3, postmeningitis in 2, congenital aqueductal stenosis in 1, hydranencephaly in 1, porencephaly in 1, and idiopathic in 5.

The mean age at surgery was 8 months (range 0.3–24 months). The mean follow-up was 64 months. In 9 cases (50%), control of hydrocephalus was successful and the patients did not require further surgeries. In 9 patients (50%), treatment failed. Of these, 3 patients underwent repeat ETV 2, 3, and 38 months after the initial surgery. Endoscopic observation of the previous coagulation site revealed no regeneration of the choroid plexus in 2 patients, who underwent repeat ETV 2 and 3 months after CPC. In 1 patient who underwent repeat ETV 38 months after CPC, no regeneration of the choroid plexus, except for that in the proximity of the foramen of Monro, was observed.

Conclusions. Endoscopic CPC with or without ETV can be a safe and effective treatment alternative to shunt placement in infantile hydrocephalus. Obliteration of the choroid plexus can persist in the relatively long term following CPC, which may contribute to the long-term control of hydrocephalus in successful cases.

Methods

We reviewed the charts of 18 consecutive patients with hydrocephalus primarily treated by endoscopic coagulation of the choroid plexus at the National Center for Child Health and Development, Tokyo, over the period of July 2002 to July 2012. Retrospective analysis for patients’ information, including age, sex, presenting symptoms, surgical treatment, and clinical outcomes, was performed using the medical charts.

Diagnosis of hydrocephalus was made on the basis of clinical manifestation including growth of head circumference, tight fontanel, vomiting, and lethargy, as well as by imaging studies revealing dilatation of ventricles. Preopera-
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tive MRI and CT were performed in all patients to detect any obstructive etiology for hydrocephalus. Cine phase-contrast MRI and/or CT cisternography was used to detect any obstruction of CSF flow including aqueductal stenosis or fourth ventricular outlet obstruction. We considered hydrocephalus to be "communicating" if there were no obstructions detected on the imaging studies and to be "obstructive" if any CSF flow obstructions were demonstrated on imaging studies. Typically, we selected ETV alone for obstructive hydrocephalus due to aqueductal stenosis or fourth ventricular outlet obstruction. We occasionally selected a combination of CPC and ETV for young infants with obstructive hydrocephalus because CSF absorption has not been fully developed in young infants. Ventriculoperitoneal shunt placement was usually selected in cases of communicating hydrocephalus. When parents strongly objected to shunt placement and desired endoscopic procedures as an initial treatment in their children, CPC or a combination of CPC and ETV was selected. Thus, 5 patients with obstructive hydrocephalus and 7 patients with communicating hydrocephalus were treated by a combination of CPC and ETV, and 6 patients with communicating hydrocephalus were treated by CPC alone.

Regarding the endoscopic CPC, a frontal approach was selected for patients who underwent concurrent ETV. A parietal approach was usually chosen for patients who underwent CPC alone. A nondominant side was usually selected. All the procedures were performed using a flexible endoscope. The outer diameter of endoscopes was 2.7, 4.4, or 4.8 mm. For a frontal approach, an entry site was made at approximately 1 cm anterior to the coronal suture and more lateral to the midpupillary line for coagulation of the contralateral side. For a parietal approach, an entry site was made at the high parietal region and 3–4 cm lateral from the midline, which would be the optimal point for access both to the foramen of Monro and the temporal horn. Using an endoscopic monopolar cautery electrode, all visible parts of the bilateral choroid plexus, from the foramen of Monro into the temporal horn, were coagulated. When the septum pellucidum was intact, a septostomy was performed in the proximity of the foramen of Monro, and the contralateral choroid plexus was coagulated.

Postoperatively, neuroimaging studies were acquired in all patients, and the size of the ventricles was assessed at 2 weeks, 6 months, and 1 year after the surgery. Thereafter, neuroimaging studies were done on a yearly basis. Patients were followed up periodically at outpatient visits. If patients’ signs and symptoms suggested increased intracranial pressure, a repeat neuroimaging study was performed at that time to assess the size of the ventricles. Treatment was deemed successful when clinical signs regressed and when no repeat surgery was needed.

We analyzed the time to treatment failure by using the Kaplan-Meier method to construct survival curves. Statistical analysis was performed using the log-rank test to compare the 2 groups. A value of p < 0.05 was considered significant.

Results

From July 2002 to July 2012, 18 patients with hydrocephalus underwent endoscopic CPC as an initial surgical treatment. None of the patients had received shunts before CPC. Nine patients were male and 9 were female, whose mean age at surgery was 8 months (range 0.3–24 months). The etiology of hydrocephalus was posthemorrhagic in 5 patients, myelomeningocele in 3, postmeningitis in 2, congenital aqueductal stenosis in 1, hydranencephaly in 1, porencephaly in 1, and idiopathic in 5. In 12 patients, ETV was concurrently performed. The etiology of hydrocephalus in those who underwent combined CPC and ETV was posthemorrhagic in 3 patients, myelomeningocele in 3, postmeningitis in 1, congenital aqueductal stenosis in 1, and idiopathic in 4. There were no perioperative complications in any patient.

The mean follow-up period was 64 months (range 17–135 months). In 9 patients (50%), hydrocephalus was successfully controlled and the patients did not require further surgeries; the mean age of these patients was 9.8 months (range 3–24 months). Of these, 4 patients underwent CPC alone and 5 patients underwent combined CPC and ETV. Postoperative imaging studies demonstrated a slight decrease in the ventricular size in all patients, with successful control of hydrocephalus.

In 9 patients (50%), treatment failed. The mean age of these 9 patients was 5.9 months (range 0.3–12 months). Of these, 2 patients underwent CPC alone and 7 patients underwent combined CPC and ETV. The median interval to treatment failure was 3 months (range 1–38 months). The survival curves in Fig. 1 show the time to treatment failure for patients who underwent CPC alone in comparison with those who underwent combined CPC and ETV. The difference was not statistically significant according to the log-rank test (p = 0.21). A ventriculoperitoneal shunt was placed in 6 patients. Three patients underwent repeat ETV 2, 3, and 38 months after the initial surgery. The etiology of hydrocephalus in these patients was posthemorrhagic, myelomeningocele, and congenital aqueductal stenosis, respectively. The ETV site was occluded in all 3 cases. Endoscopic observation of the site of previous coagulation, which covered the entire choroid plexus site, including the temporal horns of both ventricles, revealed no regeneration of the choroid plexus in 2 patients who underwent repeat ETV 2 and 3 months after CPC. In 1 patient who underwent repeat ETV 38 months after CPC, no regeneration of the choroid plexus, except for that in the proximity of the foramen of Monro, was observed (Fig. 2).

Discussion

The efficacy of CPC with or without ETV has been demonstrated for selected patients with hydrocephalus.1,4–11 Pople and Ettles wrote that the success of CPC was influenced by the type and the rate of progression of hydrocephalus.6 They reported that CPC offered a much higher rate of control in patients with communicating hydrocephalus than in those with obstructive hydrocephalus: 38% (13 of 34) and 11% (2 of 18), respectively. They also reported a higher rate of control in patients with a full or normal anterior fontanel than in those with a tense fontanel: 46% (19 of 41) and 13% (2 of 16), respectively. Based on these data, the authors indicated that patients with slowly progressive
communicating hydrocephalus without tense fontanels were good candidates for CPC. They stated the rationale for using CPC to treat hydrocephalus is that reduction in CSF restores the balance of production and absorption of CSF and halts the progression of the hydrocephalus in selected patients. Warf et al. reported that CPC in combination with ETV is useful for hydrocephalus in young infants. The etiologies of the hydrocephalus that they treated included postinfectious, posthemorrhagic, myelomeningocele, Dandy-Walker complex, and congenital aqueductal stenosis. They postulated that both obstructive and communicating components of hydrocephalus could be treated by using the combined treatments. Thus, the main indication for CPC alone as the first-line treatment is considered to be slowly progressive communicating hydrocephalus, and the indication for CPC in combination with ETV is considered to be a young infant with obstructive hydrocephalus.

We also performed a combination of CPC and ETV in 7 patients with communicating hydrocephalus. The rationale for performing ETV in addition to CPC for communicating hydrocephalus was that ETV can be effective for communicating hydrocephalus by venting ventricular CSF through the stoma, reducing the increased systolic pressure, and inhibiting an enlargement of ventricles as previous studies have suggested. The combined CPC and ETV approach was successful in 2 of the 7 patients with communicating hydrocephalus in our series.

Several reports have demonstrated the long-term efficacy of CPC with or without ETV, with a mean follow-up of approximately 10 years. However, the question of whether the effect of the coagulation is temporary and the choroid plexus regenerates, but in the meantime CSF absorption developed further, or whether the choroid plexus is obliterated by the coagulation and the long-term control of hydrocephalus is achieved by this obliteration remains unanswered. In our series, by direct endoscopic inspection, we observed 3 cases of total or near-total obliteration of the choroid plexus after coagulation was performed. Endoscopic inspection of the previous coagulation site revealed no regeneration of the choroid plexus in 2 patients who underwent repeat ETV 2 and 3 months after CPC. In 1 patient who underwent repeat ETV 38 months after CPC, no regeneration of the choroid plexus occurred except for a portion in the proximity of the foramen of Monro. These results indicate that choroid plexus obliteration can persist in the relatively long term and may contribute to the long-term control of hydrocephalus in successful cases of CPC.

In 1 patient, choroid plexus regeneration was observed in the proximity of the foramen of Monro 38 months after CPC. This occurrence may be due to incomplete coagulation of the blood vessels within the choroid plexus resulting from an attempt to preserve the thalamostriate vein. It is important for achieving obliteration of the choroid plexus to coagulate not only the choroid plexus but also all the blood vessels within the choroid plexus, including the feeders from the anterior choroidal artery and the lateral posterior choroidal artery and the superior choroidal vein, until they turn white and shrivel up, as previous authors have pointed out.

Hallaert et al. have also reported the long-term obliteration of the choroid plexus after CPC. They performed CPC in a patient with choroid plexus hyperplasia and reported that MRI studies obtained 3 years later showed no residual enlargement of the choroid plexus, although they did not inspect the structure directly using an endoscope. They had performed coagulation until the bulky reddish fronds of the choroid plexus faded into a grayish shriveled

Fig. 1. Graph showing the time to treatment failure of the 2 patient groups. The dotted line represents patients treated with CPC alone, and the solid line represents patients treated with combined CPC and ETV.

Fig. 2. Intraoperative endoscopic view at the initial operation showing the right lateral ventricle before (A) and after (B) CPC. Intraoperative endoscopic view at repeat ETV 38 months after the initial surgery demonstrating no regeneration of the choroid plexus (arrowheads) except for that in the proximity of the foramen of Monro (arrow, C).
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mass. They also advocated that both the vascular supply and the fronds of the choroid plexus itself should be coagulated to achieve choroid plexus obliteration. According to their report and ours, obliteration of the choroid plexus can persist in the relatively long term following CPC. It is thought that CPC may control hydrocephalus in the long term in selected cases by obliterating the choroid plexus, reducing CSF production, and maintaining the balance between production and absorption of CSF.

Conclusions

Choroid plexus coagulation with or without ETV can be a safe and effective treatment alternative to shunt placement in infantile hydrocephalus. Obliteration of the choroid plexus can persist in the relatively long term following CPC, which may contribute to the long-term control of hydrocephalus in successful cases.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: all authors. Acquisition of data: Ogiwara, Uematsu. Analysis and interpretation of data: all authors. Drafting the article: Ogiwara. Critically revising the article: Morota. Reviewed submitted version of manuscript: Ogiwara. Approved the final version of the manuscript on behalf of all authors: Ogiwara. Statistical analysis: Ogiwara.

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