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Endoscopic Treatment Strategy for a Disproportionately Large Communicating Fourth Ventricle: Case Series and Literature Review

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Abstract

An isolated fourth ventricle (IFV) is characterized by fourth ventricular dilation due to obstruction of its inlet and outlet. A disproportionately large communicating fourth ventricle (DLCFV) is a rare subtype of IFV, characterized by dilation of the fourth ventricle, regardless of the size of the lateral ventricles, with no apparent obstruction of the cerebral aqueduct. To our knowledge, this is the first case series describing endoscopic diagnosis and treatment strategy for DLCFV. We retrospectively reviewed six cases of DLCFV in which endoscopic surgery was performed at our institution and affiliated facilities between June 2013 and March 2017. DLCFV was diagnosed using radiographic imaging and intraoperative endoscopy. We also conducted a PubMed search and included only original studies related to DLCFV treatment written in English in our review of the literature. Endoscopic third ventriculostomy (ETV) was performed in all patients. Additional endoscope-assisted placement of a fourth ventriculoperitoneal (VP) shunt was performed in two patients who could not be managed with ETV alone because of severe adhesion of the interpeduncular cistern due to subarachnoid hemorrhage (SAH). The patients' symptoms and the size of the fourth ventricle improved with surgical treatment, without complications. Endoscopic surgery for DLCFV appears to be a safe and effective treatment. Based on our treatment strategy, ETV is the first-line treatment for DLCFV. Endoscope-assisted placement of the fourth VP shunt can be treatment for severe adhesion of the interpeduncular cistern.

Key words: endoscopic third ventriculostomy, hydrocephalus, isolated fourth ventricle, ventriculoperitoneal shunt

Introduction

A disproportionately large communicating fourth ventricle (DLCFV) is a rare subtype of an isolated fourth ventricle (IFV) that is a type of obstructive hydrocephalus, although IFV occurring after infections or shunt placement has been previously reported and discussed.[1–5] IFV develops as a marked dilation of the fourth ventricle due to the obstruction of its outlet and the aqueduct. In contrast, DLCFV, although a subtype of IFV, is characterized by apparent patency of the aqueduct.[6] Since the first report of DLCFV in 1980,[4] a limited number of cases have been reported. The standard treatment for DLCFV has not been determined, although several procedures to treat occlusive hydrocephalus are well established. Treatments for occlusive hydrocephalus include ventricular drainage, ventriculoperitoneal (VP) shunt, lowering of the opening pressure of the adjustable valve, endoscopic third ventriculostomy (ETV), aqueductoplasty with or without stent placement, interventriculostomy, septostomy, and foraminoplasty.[6–10] Some of the reported cases of DLCFV developed after ventricular drainage or VP shunt placement for hydrocephalus.[11] DLCFV differs from other occlusive hydrocephalus in this respect. The root cause of DLCFV is obstruction of the outlet of the fourth ventricle, but the

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pathophysiology has not been well described so far. Endoscopic treatment could be the first-line treatment for DLCFV because DLCFV is one of the types of obstructive hydrocephalus. In addition, endoscopic findings could help clarify the pathophysiology of DLCFV. One study showed that ETV was effective for a patient with DLCFV.[6] We routinely performed endoscopic surgery for DLCFV based on the first case report of the usefulness of ETV in the patient with DLCFV. Here, we present the first case series of endoscopic diagnosis and surgical treatment of patients with DLCFV. We describe the endoscopic surgery and treatment strategy for DLCFV and review the literature.

Materials and Methods

In this retrospective study, all six patients with DLCFV who underwent endoscopic surgeries at Nagoya University and affiliated facilities between June 2013 and March 2017 were included. Written informed consent was obtained from all individual participants included in the study. We diagnosed DLCFV using radiographic imaging and intraoperative endoscopy that revealed an enlargement of the entire cerebral ventricular system with a particularly marked dilatation of the fourth ventricle and patency of the aqueduct. Hence, patients whose radiographic imaging showed equal enlargement of the entire cerebral ventricular system were excluded. Moreover, patients who did not undergo surgery were excluded from this study.

According to our strategy, ETV is the first-line treatment for DLCFV. The operative method of ETV was standard under the direct view of the flexible endoscope: we fenestrated the floor of the third ventricle at the tuber cinereum between both mammillary bodies and the infundibular recess with a balloon catheter. Endoscope-assisted placement of a fourth VP shunt could be an additional treatment in patients with a severe adhesion of the interpeduncular cistern. The operative method of endoscopic-assisted placement of a fourth VP shunt is as follows: first, we insert a ventricular catheter into the lateral ventricle. Second, we place the ventricular catheter sequentially into the foramen of Monro, the cerebral aqueduct, and finally, the fourth ventricle, while investigating the structures under the direct view of the flexible endoscope.[11]

Literature review

For this study, PubMed searches for the terms "disproportionately large communicating fourth ventricle," "isolated fourth ventricle," and "fourth ventricular outlet obstruction," combined with a "human species" filter were performed. Only original studies related to DLCFV treatment written in English were included. We excluded studies that could not definitively diagnose DLCFV and that did not document treatment for the condition. Hence, a recently published case series was excluded from this review because it did not mention the characteristics or provide the images of each patient.[12]

Results

Six patients with DLCFV were identified during the study period. A summary of the patient characteristics is shown in Table 1. The mean duration of the follow-up period was 44.7 months (range, 32–60). The mean age of the patients was 38.5 years (range, 2–67), and three of them were women. Three patients had no particular medical history other than hydrocephalus, although a medical history of hydrocephalus resulted from subarachnoid hemorrhage (SAH) in two patients and from intraventricular hemorrhage (IVH) in one patient. Four patients received ventricular drainage or VP shunt as the initial treatment for hydrocephalus, but the fourth ventricular dilatation did not resolve.

All patients had patency of the aqueduct based on the intraoperative endoscopic observation. All patients underwent ETV. Two patients, who had a severe adhesion of the interpeduncular cistern due to SAH, underwent an additional endoscope-assisted placement of a fourth VP shunt to treat the hydrocephalus. Using endoscopy, we could accurately place the ventricular catheter in the fourth ventricle through the cerebral aqueduct. All patient symptoms improved after ETV or additional endoscope-assisted placement of a fourth VP shunt, and all procedures were complication-free. The size of the fourth ventricle improved to varying degrees via surgical treatment in all patients.

Illustrated cases

Case 1 and Case 4 are described in detail, as they are representative of the six patients we treated for DLCFV. Case 1 had no particular medical history related to hydrocephalus, whereas Case 4 had a history of intracranial hemorrhage.

Case 1

A 43-year-old man presented with nausea and ataxic gait disturbance. The findings of a systemic examination and the patient's medical history were normal. Computed tomography (CT) revealed that the cerebral ventricles were enlarged; a ventricular drainage tube had already been placed in a previous hospital (Figs. 1A and 1D). He was referred to us

Table 1 Patient characteristics from six endoscopically treated cases of disproportionately large communicating fourth ventricle

Case number	Age (years)/sex	Etiology	Operative history	Aqueduct	Adhesion of the basilar cistern	Treatment	Outcome	Follow-up (months)
1	43, M	Idiopathic	Ventricular drainage	Patent	(–)	ETV	Improvement	52
2	67, F	Idiopathic	None	Patent	(–)	ETV	Improvement	40
3	46, F	Idiopathic	None	Patent	(–)	ETV	Improvement	32
4	39, F	SAH	VP shunt	Patent	(+)	ETV, placement of a fourth VP shunt	Improvement	60
5	34, M	SAH	VP shunt	Patent	(+)	ETV, placement of a fourth VP shunt	Improvement	51
6	2, M	IVH	Ventricular drainage	Patent	(–)	ETV	Improvement	33

ETV: endoscopic third ventriculostomy, F: female, IVH: intraventricular hemorrhage, SAH: subarachnoid hemorrhage, M: male, VP: ventriculoperitoneal.

for additional treatment because his symptoms and the size of the fourth ventricle did not improve within 2 weeks after the first surgery. Postoperative images revealed enlargement of the entire ventricular system with particularly marked dilatation of the fourth ventricle (Figs. 1B and 1E). Thus, we diagnosed the patient with DLCFV and performed ETV. We could not detect any indication of hemorrhage, infection, or other abnormality in the ventricle. Postoperatively, the size of the fourth ventricle decreased (Figs 1C and 1F). The patient's ataxia was relieved gradually 2 weeks postoperatively.

Case 4

A 39-year-old woman presented with a headache. She was transferred to our hospital after a CT scan indicated the presence of SAH due to a ruptured aneurysm in the right posterior inferior cerebellar artery. We clipped the aneurysm and performed ventricular drainage. We tentatively diagnosed her with hydrocephalus caused by occlusion of the foramina of Luschka and Magendie and placed a VP shunt. Following the gradual decrease in the valve pressure to 80 mmH_2O because of enlargement of the fourth ventricle, the patient developed an acute subdural hematoma. We removed the hematoma and performed decompression. Subsequently, we adjusted the shunt valve pressure from 80 to 160 mmH_2O and gradually reduced it to 120 mmH_2O. The size of the lateral and third ventricles decreased, but the fourth ventricle remained enlarged (Figure 2A). Magnetic resonance imaging (MRI) and cine MRI indicated patency of the cerebral aqueduct (Figs. 2B and 2C). We diagnosed the patient with DLCFV based on these imaging findings and performed ETV. Endoscopic operative findings indicated a dilated cerebral aqueduct and thickening of the bottom wall of the third ventricle (Figs. 2D and 2E), and the interpeduncular cistern showed adhesion resulting in restricted cerebrospinal fluid (CSF) flow. After the first operation, CT-cisternography revealed that the contrast agent was stagnated in the fourth ventricle, and the symptoms of the patient did not improve. We performed endoscope-assisted placement of a fourth VP shunt. After the second endoscopic operation, the size of the fourth ventricle decreased without the presence of slit lateral ventricles within 2 weeks, and the patient's symptoms were relieved gradually (Figs. 2F and 2G).

Literature review

We identified 13 case reports that described 21 patients and focused on DLCFV treatment. A summary of the patient characteristics is shown in Table 2. The patients comprised 8 women and 13 men (mean

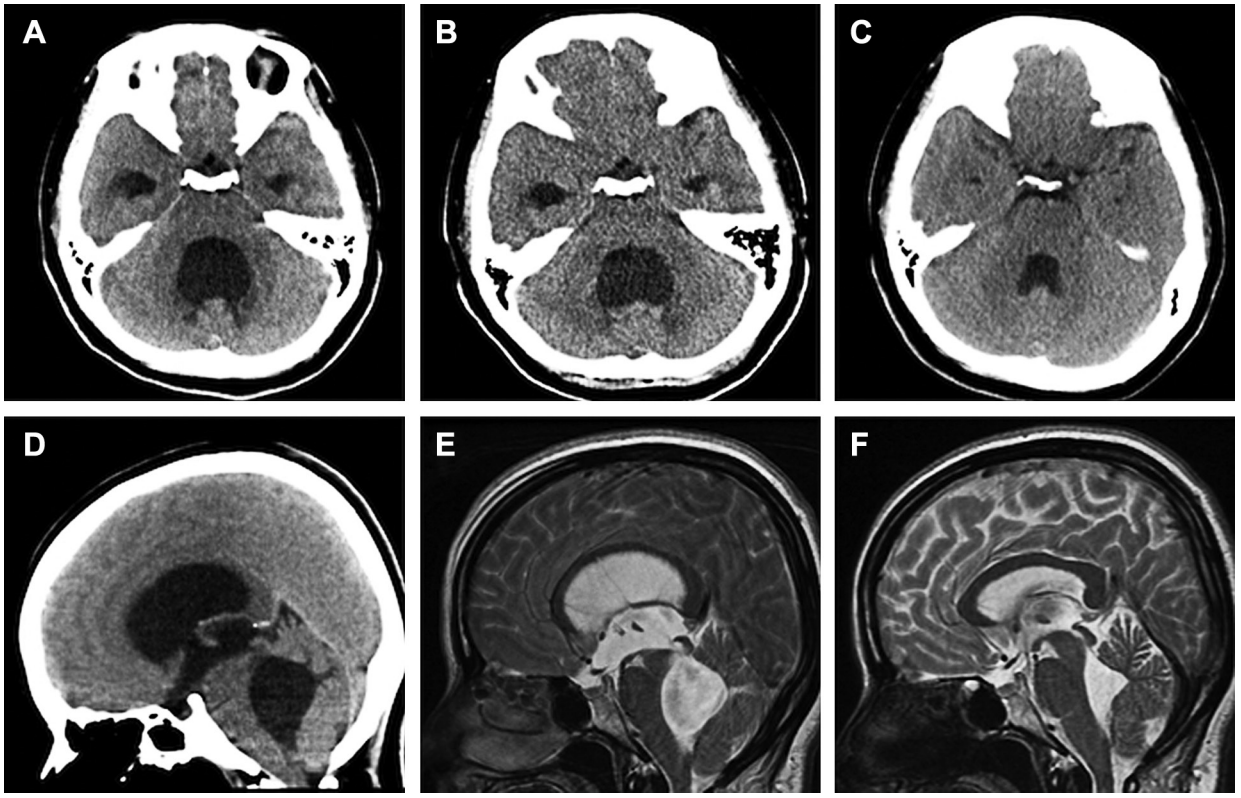

Fig. 1 Preoperative computed tomography scans (A and D), postoperative computed tomography scans (B and C), and postoperative T2-weighted magnetic resonance images (E and F) of Case 1. Axial (A, B, and C) and sagittal (D, E, and F) images. Postoperative images after placement of a ventricular drainage tube (B and E) showing no improvement of enlargement of the entire ventricular system with particularly marked dilatation of the fourth ventricle. Postoperative images after the endoscopic surgery (C and F) showing that the size of the fourth ventricle has decreased.

age, 29.6 years). In all, 13 patients had no medical history related to hydrocephalus, whereas five patients had a history of intracranial hemorrhage. The other three patients had a history of meningitis, pilocytic astrocytoma, and myelomeningocele. Nine patients had a surgical history, including VP shunt, ETV, ventricular drainage, and Ommaya reservoir placement. All patients underwent preoperative CT or MRI, which revealed the characteristic imaging of DLCFV. In these reports, the methods of treatment for DLCFV (ETV in seven patients, placement of a fourth VP shunt in six patients, suboccipital craniectomy or craniotomy for excision of the outlet membrane in three patients, suboccipital craniectomy with fourth ventricular fenestration, and partial resection of the tonsil in one patient) were mentioned. Four of six patients who had an operative history of VP shunting underwent placement of a fourth VP shunt. Five patients had a history of intracranial hemorrhage and underwent surgery for a fourth VP shunt. Patient symptoms were improved in all cases without complications, as summarized in Table 2.

Discussion

Diagnosis

DLCFV is characterized by the dilation of the fourth ventricle regardless of the size of the lateral ventricles and patency of the inlet of the fourth ventricle, whereas most cases of hydrocephalus are demonstrated by the dilation of the cerebral ventricles. We revealed the disproportionately large fourth ventricle and patency of cerebral aqueduct on radiographic images and upon endoscopy in this study. We considered that the endoscopic operative findings contributed to the definitive diagnosis of DLCFV.

Mechanisms

Various mechanisms underlying the development of DLCFV have been proposed in previous studies.[13–15] These studies concluded that when the occlusion is in or near the fourth ventricular outlet, pressure in the fourth ventricle likely results from the vulnerability created by CSF pulse waves and the water

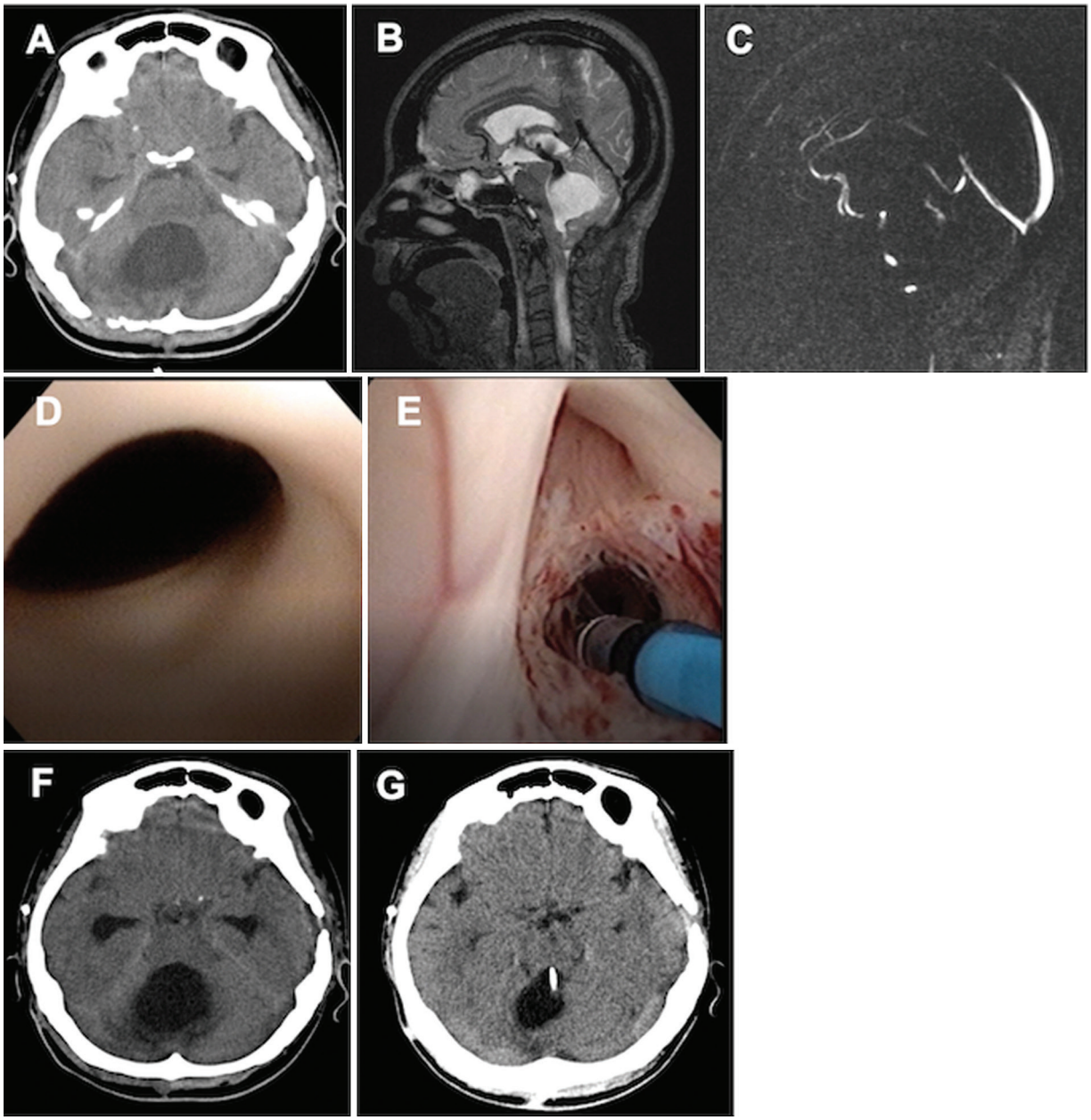

Fig. 2 Preoperative images of the third ventriculostomy before endoscopy. Computed tomography (A, F, and G) and sagittal T2-weighted magnetic resonance imaging (B) of a patient (Case 4) showing the reduced size of the lateral and third ventricles and the unchanged size of the fourth ventricle. Magnetic resonance images also show tonsillar herniation and syringomyelia. Preoperative cine magnetic resonance imaging (C) showing patency of the cerebral aqueduct. Endoscopic findings indicating the dilated cerebral aqueduct (D) and the thickening of the bottom wall of the third ventricle (E). Preoperative (F) and postoperative (G) images after endoscope-assisted placement of a fourth ventriculoperitoneal shunt, showing that the size of the fourth ventricle decreased after the operation.

hammer effect. Thus, ventricular drainage or VP shunt is not a definitive treatment for this condition. Moreover, these surgical treatments, including ventricular drainage or VP shunt, might worsen the condition with DLCFV due to supratentorial overdrainage.[11]

In our study, two of the six patients had SAH and one of the six patients had IVH. In our literature review, five of the 21 patients had a history of intracranial hemorrhage and three of the 21 patients had a history of meningitis, myelomeningocele, and pilocytic astrocytoma. These findings suggested that

Table 2 Literature review of reported cases of disproportionately large communicating fourth ventricle

Author (Year)	No. of patients	Age (years)/sex	Etiology	Operative history	Treatment	Outcome
Zimmerman et al. (1978)[5]	1	43, M	Idiopathic	None	VP shunt	Improvement
Rifkinson-mann et al. (1987)[22]	1	42, M	Idiopathic	None	Suboccipital craniectomy excision of the outlet membrane	Improvement
	2	52, M	Idiopathic	None	Suboccipital craniectomy excision of the outlet membrane	Improvement
Mohanty et al. (1999)[21]	1	32, M	Idiopathic	None	ETV	Improvement
	2	20, F	Idiopathic	None	ETV	Improvement
	3	45, F	Idiopathic	None	ETV	Improvement
Huang et al. (2001)[18]	1	15, F	Idiopathic	None	Suboccipital craniotomy excision of the outlet membrane	Improvement
Karachi et al. (2003)[19]	1	21, F	Idiopathic	None	ETV	Improvement
	2	53, F	Idiopathic	None	ETV	Improvement
	3	68, M	Idiopathic	None	ETV	Improvement
Longatti et al. (2006)[20]	1	64, F	Idiopathic	None	Opening of the foramen of Magendie	Improvement
Hagihara et al. (2007)[16]	1	13, M	Idiopathic	None	VP shunt	Improvement
Hashimoto et al. (2014)[17]	1	1, M	Idiopathic	Ventricular drainage	ETV	Improvement
Sartoretti-Schefer et al. (2000)[4]	1	19, M	Meningitis	VP shunt	Suboccipital craniectomy fourth ventricular fenestration and partial resection of the tonsil	Improvement
Shin et al. (2000)[10]	1	36, M	Pilocytic astrocytoma	VP shunt	Placement of a fourth VP shunt	Improvement
Yamashita et al. (2012)[25]	1	44, F	SAH	VP shunt	Placement of a fourth VP shunt	Improvement
Katano et al. (2012)[23]	1	6 wk, F	Myelomeningocele	VP shunt	Shunt valve adjust	Improvement
Ogiwara et al. (2013)[11]	1	13, M	IVH	ETV	Placement of a fourth VP shunt	Improvement
	2	3, M	IVH	Ommaya reservoir	Placement of a fourth VP shunt	Improvement
	3	7, M	IVH	VP shunt	Placement of a fourth VP shunt	Improvement
	4	31, M	IVH	VP shunt	Placement of a fourth VP shunt	Improvement

ETV: endoscopic third ventriculostomy, F: female, IVH: intraventricular hemorrhage, M: male, No.: number, SAH: subarachnoid hemorrhage, VP: ventriculoperitoneal.

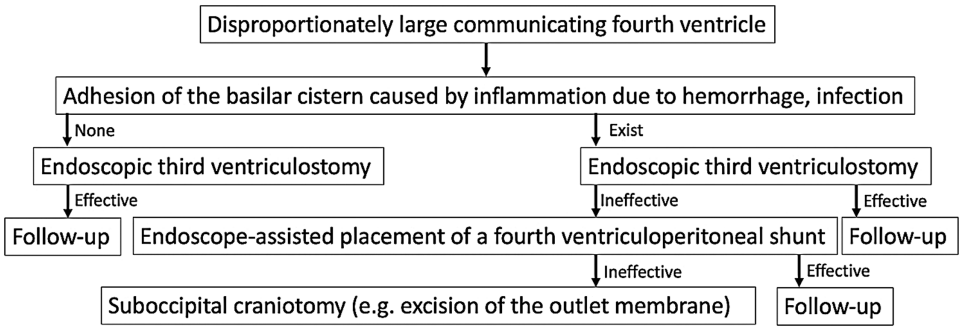

Fig. 3 Treatment algorithm for a disproportionately large communicating fourth ventricle.

DLCFV could result from adhesion after an intracranial hemorrhage, infection, congenital anomalies, or a tumor. We considered that the adhesion in the outlet of the fourth ventricle and the water hammer effect on the wall of the fourth ventricle promoted DLCFV.

However, patients without any of the abovementioned histories presented with DLCFV,[5,16–22] as seen in three of the six patients in our study. Moreover, 13 of the 21 patients had no medical history related to hydrocephalus in our review of the literature. The mechanism of DLCFV in these patients was not detected, but membranous obstruction of the fourth ventricle outlet was detected by MRI and/or endoscopic findings in all patients of our case series. We hypothesized that the minor adhesion in or near the outlet of the fourth ventricle caused by inflammation due to infection, minor hemorrhage might have influenced the development of DLCFV in the patients who had no medical history related to hydrocephalus.

Treatment for DLCFV

We must understand the possibility that conventional treatments for hydrocephalus such as ventricular drainage or VP shunt may lead to the expansion of the fourth ventricle and shrinkage of the lateral and third ventricles, as discussed above.[10,11,23–25] In the literature, ETV or placement of a fourth VP shunt was performed in 13 of 21 patients. Contrarily, we performed ETV as the first-line treatment for all patients with DLCFV. We can predict the outcome of ETV based on the Endoscopic Third Ventriculostomy Success Score (ETVSS).[26] The etiology of obstructive hydrocephalus is one of the significant factors that influence the outcome of ETV. Similar to obstructive hydrocephalus due to aqueduct stenosis, ETV could be an effective procedure for DLCFV, and the efficacy of ETV could be predicted with the ETVSS. The patients without adhesion in the interpeduncular cistern could be treated with ETV alone, and the patients with adhesion due to a history of SAH could not be treated with ETV alone in our case series. In such ineffective cases of ETV, we performed additional endoscope-assisted placement of a fourth VP shunt. All patient symptoms improved after ETV or endoscope-assisted placement of a fourth VP shunt without complications. Hence, these procedures were considered feasible and useful in patients with DLCFV to relieve CSF pulse waves and the water hammer effect.

Treatment algorithm for DLCFV

The standard strategy for DLCFV has not been determined yet. Our treatment algorithm for DLCFV was that ETV should be the first-line treatment because it is simple, effective, and safe, contributing to the improvement of most hydrocephalus symptoms. Initial treatment with ETV could reduce the requirement of a shunt placement and the related complications, including infection and shunt dysfunction. Endoscope-assisted placement of a fourth VP shunt could be an additional treatment when ETV is insufficient because of impaired absorption of CSF in the ventricle and adhesion of the interpeduncular cistern due to various histories, such as intracranial hemorrhage or infection. In cases where endoscopic treatment could not improve the symptoms of a patient with DLCFV, suboccipital craniotomy and excision of the outlet membrane could be considered as the next treatment option. Our treatment algorithm for DLCFV is presented in Fig. 3.

Complications following endoscopic treatment

Endoscopic treatment may occasionally lead to complications, including infection, hemorrhage, or cranial nerve palsy.[10,27] Kulkarni et al.[28] demonstrated complications of ETV and suggested that the most common postoperative complications were CSF leak (4.4%), hyponatremia (3.9%), and pseudomeningocele

(3.9%). Contrarily, according to a meta-analysis of previous studies on aqueductoplasty or stent placement in the cerebral aqueduct, transient and permanent disconjugate eye movements were reported in 2%–7% of cases.[11,29] Ogiwara et al.[11] reported that perioperative complications of aqueductoplasty were subdural hygroma, transient disconjugate ocular movements, and transient third nerve palsy. Our intraoperative endoscopic findings confirmed that the cerebral aqueduct of patients with DLCFV was relatively expanded compared with that of patients with common hydrocephalus. This simplified the placement of a shunt catheter in the fourth ventricle through the expanded aqueduct. Therefore, the fourth VP shunt for the DLCFV patient seemed to have a lower complication rate compared to the other types of hydrocephalus. In fact, no complication was associated with the placement of the shunt catheter through the aqueduct in our series. However, the fourth VP shunt could have a relatively higher complication rate compared to ETV. Hence, we consider ETV the first-line treatment; the fourth VP shunt could be applied for patients who cannot be treated via ETV.

Limitations

The limitations of our study include difficulties associated with investigating the clinical features and outcomes of a rare disease. The inclusion of patients from previous reports may have introduced selection bias and patient heterogeneity into the study population. The efficacy of specific treatments was not established because of the very small sample size. Evaluation of each treatment with sufficient sample sizes will be required to establish its efficacy.

Conclusions

We performed endoscopic surgery in six cases of DLCFV. Endoscopic surgery is an effective, safe, and simple treatment option in patients with DLCFV. Based on our strategy, ETV is the first treatment for DLCFV. Endoscope-assisted placement of a fourth VP shunt can be a treatment for patients with severe adhesion of the interpeduncular cistern.

Conflicts of Interest Disclosure

All authors have no conflicts of interest.

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