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Case Report

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Endoscopic third ventriculostomy for patients with Blake's pouch cyst with adult-onset hydrocephalus: Importance of improved cerebrospinal fluid flow in the prepontine cistern – A case report

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ABSTRACT

Background: Blake's pouch cyst (BPC) is a posterior fossa cystic malformation that commonly occurs in children with rare adult onset. Herein, we report a case of adult onsets BPC.

Case Description: A 61-year-old man presented with gait and cognitive disturbance. Preoperative magnetic resonance imaging (MRI) revealed scarring in the prepontine cistern, and cine phase-contrast MRI revealed no pulsation. Endoscopic third ventriculostomy (ETV) was performed with opening the scarring in the prepontine cistern. Postoperative cine phase-contrast MRI revealed that cerebrospinal fluid (CSF) flow in the prepontine cistern improved, resolving the patient's symptoms.

Conclusion: We report a case of adult-onset BPC. The mechanism by which is becomes symptomatic is still unclear. We opened the scar in preportine cistern in addition to ETV with good results. In this report, we discussed the importance of the improvement in CSF dynamics in the preportine cistern.

Keywords: Adult-onset, Blake's pouch cyst, Endoscopic third ventriculostomy, Hydrocephalus, Prepontine cistern

INTRODUCTION

Blake's pouch cyst (BPC) is a cystic malformation of the posterior fossa that is characterized by the ballooning of the superior velum into the cisterna magna. BPC is often associated with hydrocephalus and commonly occurs in children but rarely in adults even if it is a congenital anomaly like a longstanding overt ventriculomegaly in adults. Herein, we report a case of BPC in a patient with adult-onset hydrocephalus who achieved good outcomes after the opening of preportine scarring as well as endoscopic third ventriculostomy (ETV).

CASE REPORT

A 61-year-old man presented with gait disturbance that worsened over 2 weeks and memory disturbance since 6 months. He had a wide-based and slow gait. His score on the timed up

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and go test (TUG) was 14.5 s and 20 steps and that on the mini-mental state examination (MMSE) was 21. Computed tomography revealed ventriculomegaly (Evans index 0.45), and magnetic resonance imaging (MRI) revealed a cystic formation at the caudal side of the cerebellum, leading to a diagnosis of BPC [Figures 1a-e]. Sagittal MRI results revealed a downward bulging third ventricle floor and membrane-like structure within the prepontine cistern [Figure 1e]. Cine phase-contrast MRI revealed a normal bidirectional flow at the level of the aqueduct canal but none at the prepontine cistern [Figures 1f and g].

We performed ETV using a flexible neuroendoscope (Olympus VEF type V[®]; Olympus, Tokyo, Japan). The third ventricle floor was opened with a perforating forceps and widened with a Expansor[®] Balloon catheter SI (Fuji Systems Corporation, Tokyo, Japan). Basilar artery and the scarring in the preportine cistern were observed through the stoma. After perforation of the prepotine scarring, we confirmed cerebrospinal fluid (CSF) flow between the third ventricle and prepontine cistern. We also opened the scarring with perforating forceps under the critical observation of a flexible neuroendoscope [Figures 1h and i]. Pulsatile movement of the third ventricular floor was more dynamic after opening the scarring compared with after ETV. During the operation, the partial lack of septum pellucidum and a very thin third ventricle floor were observed. There was no aqueduct stenosis.

Postoperative MRI indicated the opening of the floor of third ventricle and scar in the prepontine cistern. Cine phasecontrast MRI revealed CSF flow in the prepontine cistern [Figure 2]. The patient's postoperative course was good, and gait disturbance improved immediately. After 1 week postoperatively, the patient had a TUG sore of 9.1 s and 15 steps. At the end of 2 months, his memory disturbance gradually improved, and his MMSE score was 26. This patient keeps good condition even at 1 year after the operation.

DISCUSSION

Dandy–Walker cyst and BPC are both congenital anomalies that originate from the rhombencephalon tegmentum. Cerebellar vermis is formed in patients with BPC but not in those with Dandy–Walker cysts.

BPC has been considered to result from a failure of regression of Blake's pouch (the rudimental fourth ventricular tela choroidea). BPC is defined as a cystic dilatation of Blake's pouch, which is left and dilated in the preliminary step of opening of foramen Magendie. The opening of the foramen Magendie is absent or incomplete, and the CSF flow at the outlet of the fourth ventricle may be impaired. In such a case, there are cases of shown dilatation of the ventricle and presenting symptoms.

BPC can be asymptomatic or can have clinical symptoms including headache or hydrocephalus. Blake's pouch is

treated using a ventriculoperitoneal shunt; however, many studies have reported the increasing use of ETV, because ETV could avoid the risks related with shunt placement such as isolated fourth ventricle, over drainage, and infection.^[11,13] However, most cases of BPC occur in children, [2,6,10] there have been some adult cases.^[3,5,6,9] In pediatric cases, the onset of symptoms mainly begins with progressive hydrocephalus, such as cranial enlargement and sunset phenomenon, and the patient's intelligence is maintained in the normal range when hydrocephalus is controlled with proper treatment. In contrast, only 11 adult cases have been reported including our case [Table 1]. Hirono reported a child case of BPC that was not present at 8 months and appeared at age of 3 years, suggesting the presence of secondary BPC.^[7] In this case, the BPC was manifested due to a brain tumor, which disseminated ventricle wall widely and obstructed the CSF. However, as many case reports, including our case, the late manifestation of BPC remains unclear. A common symptom includes headache at a young age and gait disturbance and/ or cognitive disturbance in elderly patients. Takeshige et al.^[9] reported seven adult cases wherein ETV was performed and follow-up period was over 5 years. Of the seven cases, two cases had failed ETVs due to the unsuccessful opening of the scarring in prepontine cistern.

The scarring in the prepontine cistern is a risk factor for failure of ETV^[4,12,13] not only in the case of BPC but also any other case of hydrocephalus in pediatric case. In adult cases of BPC, in addition to ETV, it is important to open the scarring of the prepontine cistern. In the present case, the scarring of the prepontine cistern was observed, and CSF flow did not revealed on cine phase-contrast MRI preoperatively, even though the CSF flow at aqueduct was showed. Intraoperatively, we confirmed that the opening the scarring improved CSF flow, and postoperative cine phase-contrast MRI also showed CSF flow in the prepontine cistern. We thought that the improvement of CSF flow in the prepontine cistern resulted a good outcome. Moreover, it was important to confirm CSF flow during the procedure as well as to notice the scarring of prepontine cistern on preoperative MRI. Although the mechanism of scar formation remains unclear and more cases are needed to elucidate it, physicians could consider the opening of the scar in the prepontine cistern an indicator for the treatment of symptomatic BPC.

The importance of improving CSF in the prepontine cistern has been recently demonstrated. CSF pulsation is crucial for CSF circulation, and one of the most prominent areas of pulsation is the prepontine cistern.^[15,16] CSF pulsatility in prepontine cistern was disrupted in patients with hydrocephalus secondary to subarachnoid hemorrhage.^[14] In pediatric cases with an enlarged cerebral ventricle, poor pulsation in the prepontine cistern can be detected using cine MRI, unlike in asymptomatic cases.^[8] Moreover, a study by



Figure 1: (a-d) Magnetic resonance image (MRI) showing an enlarged ventricle and cystic formation at the caudal side of the cerebellum. (e) The choroid plexus extends into the cyst compatible diagnosed as Blake's pouch cyst (arrow). Sagittal MRI showing a downward bulging third ventricle floor and membrane like structure within the prepontine cistern (arrowheads). (f and g) Cine phase-contrast MRI revealing a normal bidirectional flow at the level of the aqueduct canal (arrowhead) but not at the prepontine cistern (arrow). (h and i) Intraoperative findings. (h) Scarring in the prepontine cistern (arrowheads) and basilar artery (arrows). (i) After opening the scar, abducens nerve (arrowheads).

Table 1: Summary of adult-onset Blake's pouch cyst.						
S. No.	Author	Year	Age	Gender	Symptom	Treatment
1.	Takeshige et al.	2021	30	F	Headache	ETV
2.	Takeshige et al.	2021	36	F	Headache	ETV
3.	Conti <i>et al</i> .	2003	37	F	Back pain, paresthesia	V-P shunt
4.	Takeshige et al.	2021	37	М	Headache	ETV
5.	Takeshige et al.	2021	40	М	Headache, cognitive disfunction	ETV
6.	Cornips et al.	2010	51	М	Incidental	follow up
7.	Takeshige et al.	2021	54	М	Headache, cognitive disfunction	ETV
8.	Takeshige et al.	2021	58	М	Cognitive disfunction, gait disturbance	ETV
9.	Takeshige et al.	2021	60	М	Headache, cognitive disfunction, gait disturbance	ETV
10.	Calabrò et al.	2000	61	F	Incidental	Follow up
11.	Calabrò et al.	2000	62	F	Headache	Follow up
12.	Cornips et al.	2010	69	М	Headache, cognitive disfunction, gait disturbance	ETV
13.	Present case	2022	61	М	Cognitive disfunction, gait disturbance	ETV
ETV: Endoscopic third ventriculostomy, F: Female, M: Male						



Figure 2: (a and b) Postoperative magnetic resonance imaging (MRI): (a) The opening of the floor of third ventricle is observed (arrow). (b) The opening of the scarring in the preportine cistern also observed (arrow). (c and d) Cine phase-contrast MRI indicating the cerebrospinal fluid flow in the preportine cistern (arrowheads).

Anik *et al.* reported that good pulsation in the prepontine cistern can be observed after ETV in successful cases of BPC in pediatric patients.^[1]

Although BPC involves embryonic dysplasia, it can turn symptomatic in some adult patients. In this case, the scar

in prepontine cistern was observed. It is possible that the formation of scar disturbed the CSF flow and pulsation, and stagnation of CSF caused hydrocephalus. Improvement of CSF flow in prepontine cistern is essential to achieve good outcomes.

CONCLUSION

BPC rarely occurs in adults. ETV is performed in symptomatic patients aged >60 years. In this case, we opened the scar in preportine cistern in addition to ETV. CSF circulation in the preportine cistern could be improved, which might result in good outcomes. Elderly patients with BPC can be symptomatic, and the opening the scar in the preportine cistern is important for their treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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