

Delayed intracerebral hemorrhage after ventriculo-peritoneal shunt procedure: two case reports and a review of literature

X.-T. WANG^{1,2}, L.-Y. ZHANG^{1,2}, H.-T. LV², J. LIU², Y.-H. XU^{1,3}

¹Graduate School of Dalian Medical University, Dalian, Liaoning, China

²The First Affiliated Hospital of Dalian Medical University, Dalian, Liaoning, China

³Health Commission of Liaoning Province, Shenyang, Liaoning, China

Abstract. – OBJECTIVE: Ventriculo-peritoneal shunt (VPS) is a commonly used procedure for treating hydrocephalus of various causes. Delayed intracerebral hemorrhage (DICH) is regarded as a very rare complication after VPS procedure, with mechanisms still indeterminate. We report two cases of this condition whereby we discuss the characteristics and potential explanations for it in a short review of literature.

CASE REPORT: Two female patients, aged 49, 76 respectively, were admitted to our hospital for hydrocephalus in the year 2021 as ordinary participants among many other patients with the same diagnosis. Unforeseeably, what made them special was DICH situations occurred after regular VPS procedures. Luckily both of them responded well to subsequent conservative treatment with no deterioration and were discharged promisingly in the end. Surprisingly, both of the valve mechanisms in these two functioned properly so far even after the ominous DICH events. Quality of life also improved a lot for them, thus we could consider the VPS surgery successful as well as the later management of the unwanted hematomas, in other words, a full recovery from DICH.

CONCLUSIONS: Only few cases or series of DICH were reported in the past decades and the mechanisms of it still lack a verdict. We intend to attribute physical vascular injury due to a closer contact between cerebral blood vessels and the VPS catheter for DICH in the younger patient, while degenerative changes of brain tissue might be the protagonist in the elder one. More discreetness should be expected in perioperative management of VPS patients, with still a long way to go to fully understand the mechanisms of DICH and prevent the complication in highest measure.

Key Words:

Delayed intracerebral hemorrhage, Hydrocephalus, Ventriculo-peritoneal shunt, Complications.

Introduction

Ventriculo-peritoneal shunt (VPS) is an effective and the most common procedure as a mainstay treatment of hydrocephalus in today's neurosurgical practice^{1,2}, which was firstly introduced by Kausch³ in 1908. Major complications of VPS such as shunt obstruction, catheter migration, infection, seizures, subdural hematoma, valve malfunction, were extensively reported^{4,5}. Nevertheless, post-operative intracerebral hemorrhage occurs at a relatively low rate while a delayed presentation of the situation is even rarer, with very few (about 50) reports to date. Given the fact that VPS is now a mature technique commonly performed every day around the globe, thus a considerable amount of it expected, delayed intracerebral hemorrhage (DICH) following VPS came to be an exceptionally rare complication accordingly with speculations about its nature in ongoing dispute. Prevalence of this unpleasant incidence ranged approximately from 0.3% to 4.0% in previous reports^{4,6-11}.

However severe it can sometimes be, the mechanisms of DICH still remain unclear stringently speaking, though remarkable efforts had been made for seeking feasible explanations. Even a precise definition of DICH in term of "delayed" was not explicitly pointed out in the past either but we usually assume an intraparenchymal hematoma at least 48 hours afterwards shunt placement fits the description, with onset time of way over two days in most reported cases. Mortality rate tends to be higher in patients received VPS treatment for hydrocephalus if a DICH diagnosis was confirmed¹², hence certainly more challenges arise for clinicians correspondingly. We hereby report two recent cases of DICH secondary to VPS in our institution and discuss the phenomenon with a literature review. The study

was approved by Ethics Committee of The First Affiliated Hospital of Dalian Medical University and the use for all detailed information acquired informed consent from the patients and their legal representatives.

Case 1

A 49-year-old female was admitted to our hospital in late February of 2021 for gradual but progressive intellectual retrogression accompanied by urinary incontinence as well as hobble, both of which appeared shortly before admission. A recent computed tomography (CT) scan from another institution showed clear signs of hydrocephalus so a decision was primarily made by the doctors to perform a VPS procedure on her after reaching consensus with the family representatives. Only about 3 months before this, the patient went under endovascular treatment of a ruptured carotid-posterior communicating artery aneurysm and was discharged after 2 weeks of treatment with an optimistic expectancy of fine recovery. Additional major medical issues include a history of hypertension more than 10 years which was actively controlled by medication all the way, and a surgical treatment record of cervical cancer 3 years ago, which was also successful. VPS was scheduled 2 days later than admission after necessary preparations during

the meantime. A routine test of the cerebrospinal fluid (CSF), which was drained out through a regular lumbar puncture, exhibited uneventfully and so are the blood tests, with a special focus but no evidence of coagulation dysfunction. The intracerebral pressure (ICP) was determined as 150 mmH₂O assuring a pattern of normal-pressure hydrocephalus (NPH) making us set a performance level of 1.5 on a Medtronic® Strata™ programmable valve mechanism. The procedure well succeeded with a solo attempt of catheter cannulation and without bleeding events worth mentioning. The intraoperative CSF leakage into the tube was also crystal clear. Shunt system functioned properly until we encountered the DICH scenario 9 days after the surgery, before which tremendous improvements of admission symptoms were observed. We ran a CT scan on the 4th day after surgery which was satisfactory, showing a lower Evans index and favorable positions of the catheter both inside the ventricle and the pelvic cavity. Be that as it may, a sudden deterioration in general state with vomiting on the morning of the 9th day after the VPS procedure made us run an emergency CT scan to find out a hematoma of non-negligible amount along the pathway of the proximal catheter, also inside the lateral ventricles, both of which are dangerous but by far the more vicious is the intraparenchymal

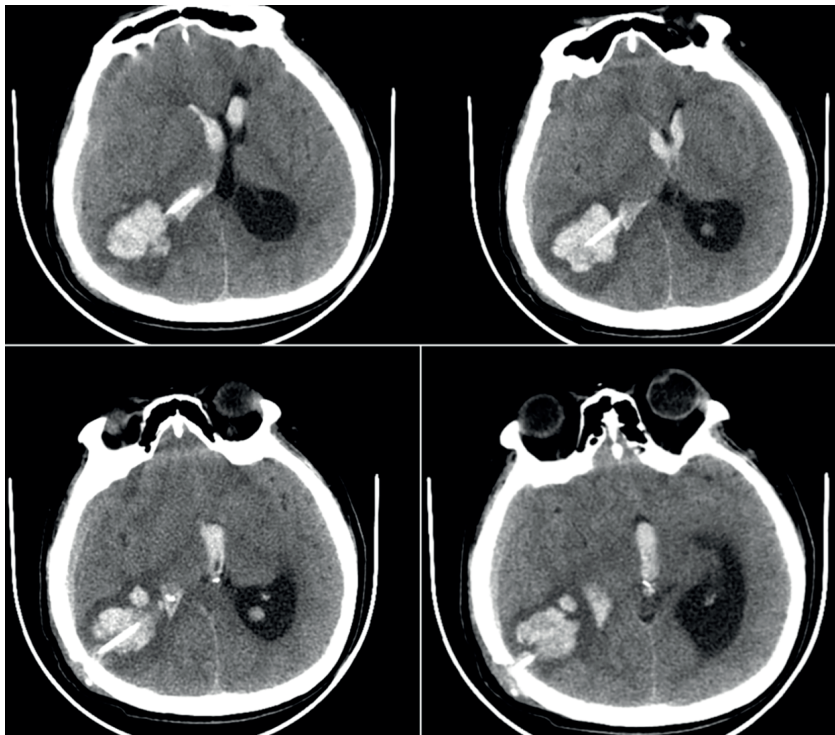


Figure 1. CT findings showing DICH of Case 1 on postoperative day 9. Hematoma was observed in right parietal lobe parenchyma and inside the ventricular system, causing subfalcial hernia. The patient was on the edge of emergency surgery for hematoma drainage and the VPS mechanism was surely endangered, but we chose conservative treatment with discretion after discussing the situation with her family. Strengthened management was active immediately with the hope to save her life firstly and reconsider the VPS system in future. CT, computed tomography; VPS, ventriculo-peritoneal shunt; DICH, delayed intracerebral hemorrhage.

one (Figure 1). Somnolence and increase in blood pressure were incorporated as simultaneous manifestations, as well as severe headache. Strengthened conservative treatment was engaged immediately since the diagnosis of DICH was basically established though unexpectedly, nonetheless, indications for surgical intervention were not ample enough. A series of CT scans showed subsequent changes of the hemorrhage (Figure 2). The patient eventually earned a stable condition with hardly potential life-threatening chances 16 days after DICH, 25 days after VPS procedure. She was then discharged for further rehabilitation in another institution for minor bradylalia and overt weakness of extremities especially in the lower limbs. Another 5 months later, we were glad to find out that the patient almost fully recovered in the follow-up. The shunt system functioned as designed by far and offered a better quality of life for her after all. Fortunately, we did not see the prognosis of this patient compromised.

Case 2

An old lady aged 76 got injured in a major car accident 6 months ago, the head trauma was conservatively treated after which she suffered from the exact “three classic signs of hydrocephalus”: instability in gait, urinary incontinence, cognitive deficits, all of which with steady deterioration. An orthopedic surgery on the right thigh was performed a couple of weeks after this very accident. Ten years ago, she was diagnosed with diabetes and this is the only highlighting of historical medical concerns. Slightly expanded lateral ventricles and interstitial brain edema were observed on an admission CT scan (Figure 3). VPS was proposed then and was performed successfully several days later on the right side. A preoperative lumbar puncture provided an initial ICP of 115 mmH₂O but nothing more of particular interests. A Medtronic® Strata™ programmable valve mechanism, initial performance level 1.0, was planted without a hitch. Another CT scan

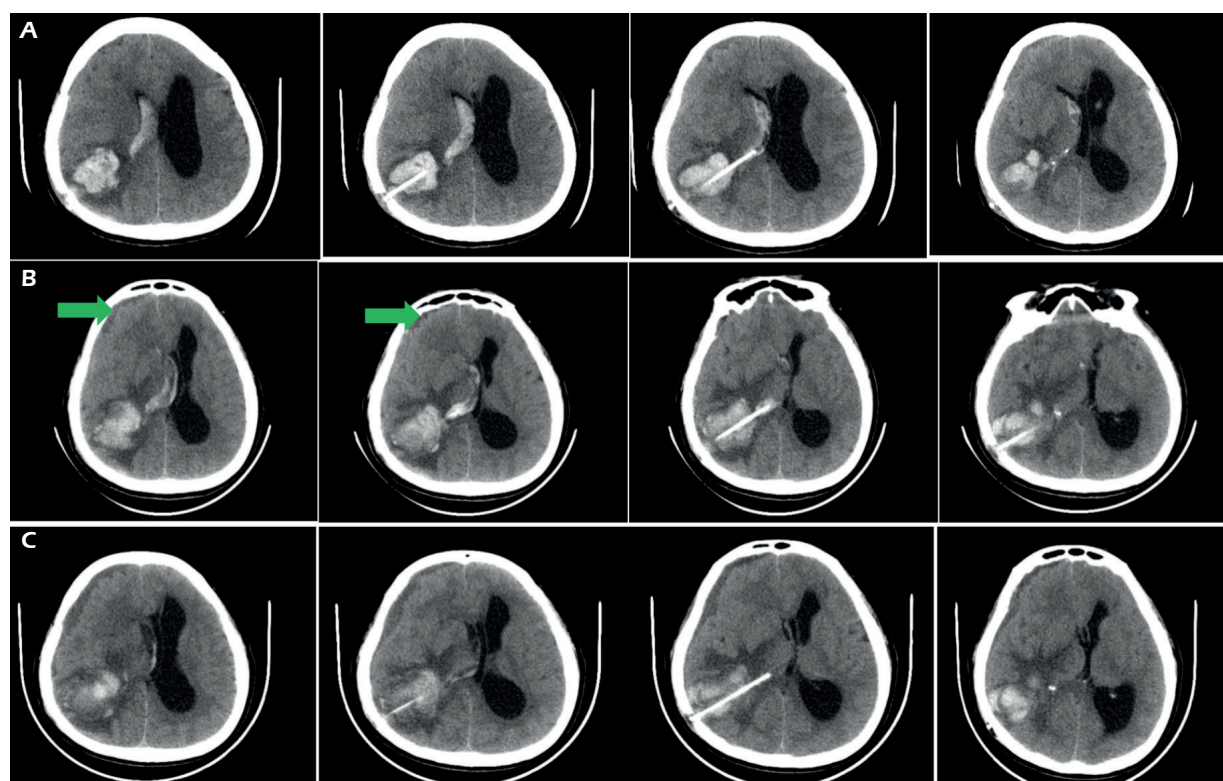


Figure 2. Serial changes of DICH on CT under conservative treatment in Case 1. Fortunately, the hematoma did not further expand, absorption changes were observed with effective conservative treatment: **A**, CT scan on postoperative day 15 (6 days after DICH), hematoma showed a similar range with onset, with the patient’s vital signs stable. **B**, CT scan on postoperative day 19 (10 days after DICH), some low-density changes were obvious in areas surrounding the hematoma, suspected slight subdural effusion (*green arrows*) was also seen possibly due to dehydration treatment. **C**, CT scan on postoperative day 25 (16 days after DICH), hematoma faded. At that time point, the patient’s life was not at risk anymore, but function of the VPS mechanism planted required follow-up. CT, computed tomography; DICH, delayed intracerebral hemorrhage; VPS, ventriculo-peritoneal shunt.



Figure 3. Admission CT scan of Case 2. It revealed signs supporting the diagnosis of hydrocephalus: ventricular expansion and interstitial brain edema. CT, computed tomography.

on the following day to VPS procedure appeared amicably (Figure 4) and the patient was in good state for the next 4 days showing credible signs of betterment. On postoperative day 6, everything seemed okay till we ran a CT scan again rather routinely after a decent interval only to find a parietal cortex hematoma flooding into the ventricle, as well as ipsilateral subdural hemorrhage (Figure 5).



Figure 4. CT scan of Case 2 on postoperative day 1. The surgery was successful, with no obstruction of CSF or hemorrhage events. This CT scan was done routinely to check on the system without any particular interests. CSF, cerebrospinal fluid; CT, computed tomography.

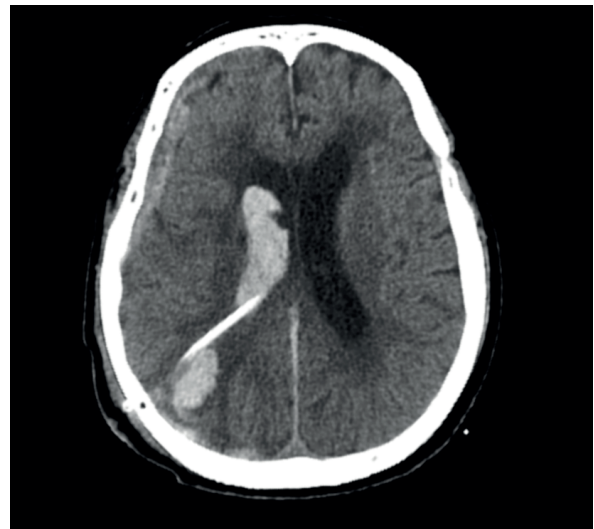


Figure 5. CT scan of Case 2 on postoperative day 6. Intraparenchymal, ventricular and subdural hematomas were observed but seemed to have mild effects because of the relatively small amount. A rare DICH situation was confirmed. This time we engaged conservative treatment with confidence. DICH, delayed intracerebral hemorrhage, CT, computed tomography.

This time a surgical approach was not applicable either out of consideration of the patient's relatively advanced age and more importantly, the mild amount of the blood. The incident was expected to have minute influence on the patient, however it went awry when a gusty seizure came up 89 hours following the most recent CT scan. We had no choice but to engage brute anti-epileptic agents in this case and fortunately it proved to be sporadic with no recurrence. No remarkable anomaly was found in CSF or blood tests during the entire perioperative period other than a minor hypokalemia that was dealt with easily. Later CT scans in the following days showed a process of bleeding absorption (Figure 6). We dismissed her another 8 days later after the seizure, on postoperative day 18. The patient, and the system, functioned well till the time of publishing.

Discussion

As a rare complication, the treatment of DICH after VPS is relatively simple because the management of the hematoma is generally consistent with other forms of brain hemorrhage, though the mortality rate can be up to 50%¹³ and for this matter, we believe this is to a large extent due to the originally catastrophic primary disorders of the central nervous system but DICH itself. In



Figure 6. CT scan of Case 2 on postoperative day 16. The patient recovered well, and the CT scan showed fine absorption of hematoma. Still, we continually worried about the status of her VPS system after this. CT, computed tomography; VPS, ventriculo-peritoneal shunt.

fact, few DICH cases subsequently undergoing forced craniotomy for hematoma evacuation have been reported whereas we believe the potentially endangered VPS system being more concerned seems appropriate. Subtle changes in protein levels of CSF could compromise the VPS tube as we traditionally consider, though challenged by some recently¹⁴, not to mention a mass of unwanted blood around the ports. We were honestly prepared for surgical removal of the valve mechanisms in our 2 cases yet enigmatically they are both fine so far to our knowledge. Still, whether DICH will increase the risk of shunt valve occlusion needs to be further studied on a large-sample basis.

Since how it came about that DICH happens such a long time after VPS surgery is a mystery, inspired by the work of Misaki et al¹⁵, we summarized the characteristics of previously reported cases in Table I, though some of them occurred within 48 hours after the procedures.

Regarding previous publications on DICH, it can be deduced that most DICH after VPS happens right around the location of catheter cannulation and (or) adjacent ventricular space. Definition of DICH mentioned above aside, moderate amount of blood inside the ipsilateral ventricle or the parenchyma near the tubing is rather common after the procedure, but mainly in the early stage^{8,13,16-18}. An incidence rate of 4% of intraparenchymal hematoma within 48 hours following VPS was reported in 1999⁸. We address the actual possibility of DICH might be greater than reported because all relevant time

duration is not constant and some small bleeding sites are very likely to be ignored for CT scans are not routinely done under many circumstances. Ma et al¹³ supported this idea with a similar statement. And there may have been some other cases of DICH unreported.

From issued literature, several possible mechanisms of DICH after VPS were elucidated:

(1) direct operational injury^{4,6,8,19}, including (A) multiple attempts on ventricular puncture during the procedure; (B) choroid plexus injury; (C) improper set of the distal tube into brain parenchyma; (2) coagulation disorder^{8,9,13}; (3) head trauma shortly after the procedure^{8,9,13}; (4) undetected vascular malformation^{8,9,13} or brain tumor^{8,13}. However we think these theories are more suitable to be described as conjectural for a preclusion use, because no much cases are reported to verify them, we hardly see a patient with brain tumor underwent VPS puncture right at site of the tumoral lesion. An exception to this may be that in some ventricular tumors causing obstructive hydrocephalus while VPS performed as a palliative strategy, the catheter goes into the ventricular system and make unavoidable contact with the tumor entities. We noticed few of the reported cases underwent a magnetic resonance imaging (MRI) or a cerebral angiography maybe because of different allocations of health care resources.

We present a “venous-stress” theory supported by multiple authors. Matsumura et al⁷ reported DICH in a 17-year-old boy on the 7th postoperative day and attributed the complication to Valsalva’s effect. The Valsalva’s effect will suppress the cerebral venous return to cause an increased cerebral venous pressure, once overloaded, leading to hemorrhage. Similarly, Misaki et al¹⁵ explained how surgical hemostasis operations on the cortical veins could cause venous return disturbances that eventually lead to hematoma formation. Venous dysfunction plays key roles in this theory.

Another plausible explanation which is more accepted by most is slow traumatic injury. The hypothesis suggests a chronic erosion of vasculature due to closer contact with the catheter blaming ventricular collapse after the shunt^{4,9,12,15}, maybe with the help of normal, cadent pulsations of the brain^{13,20}, eventually lead to DICH. However, Misaki et al¹⁵ inclined to think such closer contact is sure to happen after VPS in almost all of the cases thus it is not convincing enough to hold it responsible for rarely seen DICH, venous occlusion due to intraoperative manipulation and

Table I. Characteristics of Reported DICH Cases Secondary to VPS.

Series	Year	Onset day	Age/gender	Puncture site	Primary disease or surgery		
Matsumura et al ⁷	1985	7	17/M	AH	Head trauma		
Snow et al ⁹	1986	5-7	43/F	AH	INPH		
Derdeyn et al ⁶	1988	0	56/M	PH	Acute subdural hematoma		
		0	73/F	AH	INPH		
Mascalchi ²²	1991	15	68/M	-	ICH		
Savitz and Bobroff ⁸	1999	2	-	PH	-		
		2	-	PH	-		
Alcazar et al ⁴	2007	6	64/F	PH	SAH		
Gong et al ¹¹	2007-2013	3	62/M	AH	ICH		
		3	64/F	PH	Head trauma		
		7	76/M	AH	SAH		
		3	50/M	AH	SAH		
		4	61/F	AH	Head trauma		
		5	67/M	AH	Meningitis		
		7	65/M	AH	-		
		4	61/M	AH	-		
		3	60/M	AH	Head trauma		
		4	53/F	PH	SAH		
		5	68/F	AH	-		
		5	61/M	AH	SAH		
		Misaki et al ¹⁵	2010	0	82/F	AH	INPH
				2	80/F	AH	ICH
7	55/M			PH	SAH		
Khandelwal et al ²⁴	2011	6-13	64/M	PH	SAH		
		22	Infant/M	PH	Occipital encephalocele with communicating hydrocephalus		
Zhou et al ¹²	2011	5	32/F	AH	INPH		
		3	58/M	AH	Head trauma		
Guo et al ¹⁰	2011-2016	3	58/F	-	Aneurysm clipping		
		3	54/M	-	ICH		
		3	61/M	-	Head trauma		
		4	61/M	-	Metastatic brain tumor		
		4	75/M	-	ICH		
		5	84/F	-	Head trauma		
		6	48/F	-	Aneurysm clipping		
		6	61/M	-	-		
		6	62/M	-	Chronic subdural hematoma		
		6	78/M	-	-		
		7	64/F	-	Aneurysm clipping		
		7	65/F	-	Brain tumor		
		7	76/F	-	ICH		
		8	66/M	-	Head trauma		
		8	69/M	-	-		
		9	57/F	-	Intracranial endovascular treatment		
9	69/M	-	-				
9	72/M	-	-				
10	33/M	-	ICH				
10	30/M	-	ICH				
Koktekir et al ²⁵	2012	7 years	7/M	-	Congenital hydrocephalus		
Okazaki et al ²⁶	2013	7	Infant/F	-	Congenital hydrocephalus		
		-	Infant/M	-	Congenital hydrocephalus		
Ma et al ¹³	2015	8	69/M	AH	Head trauma		
Coulibaly et al ²³	2015	11 months	13/M	PH	INPH		
Musali et al ²⁰	2019	7	56/F	PH	-		
Our cases	2021	9	49/F	PH	SAH		
		6	76/F	PH	Trauma		

-: Data Unavailable. DICH: Delayed intracerebral hemorrhage; VPS: Ventriculo-peritoneal shunt; M: Male; F: Female; AH: Anterior horn (of lateral ventricle); PH: Posterior horn (of lateral ventricle); INPH: Idiopathic normal pressure hydrocephalus; ICH: Intracerebral hemorrhage; SAH: Subarachnoid hemorrhage.

the vulnerability of brain tissue induced by a primary brain disease were stated to be the reasons for early and delayed hemorrhage respectively in the study, which brings us to one more point of view, the fragility theory. Potential degenerative vasculopathy and other changes of the brain after severe head trauma or a craniotomy may have increased the possibility of a microvessel rupture near the catheter¹³. Admittedly, brain fragility is a relatively abstract concept which is difficult to detect on clinical images, but the theory is still indirectly supported by certain experimental studies²¹, identifying some fragile arteries surrounding microbleeds sites.

Mascalchi²² presented a patient with intraparenchymal DICH 15 days after VPS and interestingly, the hematoma was located in the contralateral hemisphere of the catheter. The author charged reduction of ICP for this incidence, believing decreased tamponade forces secondary to the shunt caused unwanted hemorrhage. Ma et al¹³ reported a case of DICH soon after lowering adjustment of the valve threshold, which seemed reasonable to consider the sudden drop of ICP (CSF overdrainage) responsible. However, Coulibaly et al²³ suggested a drop of ICP was the cause of subdural or epidural hematoma after VPS, by no means intraparenchymal.

Moreover, Khandelwal et al²⁴ presented a rare case of bilateral thalamic hemorrhage after VPS, they hypothesized that mechanical injury to the brain causes the release of thromboplastin into the circulation activating the development of disseminated intravascular coagulation (DIC), which also seems a possible cause of DICH secondary to VPS for this matter.

Additionally, based on the clinical experience of intracerebral hemorrhage (ICH), hypertensive or not, we are confident that cerebral amyloid angiopathy (CAA) contributes to DICH secondary to VPS in elder patients since CAA is a common condition when aged and we truly have seen many cases with CAA-related ICH diagnosis, and similarly, it probably goes the same for postoperative subdural hematoma of VPS because elder patients usually have enlarged subdural space with natural encephalopathy. This reached consensus with the work by Guo et al¹⁰.

In 2012, Koktekir et al²⁵ reported a case of DICH in a 7-year-old boy who had VPS treatment, presuming the cause may be stretching of the shunt tube at the neck during the growth that leads to brain contusion. Which not be relevant in adult DICH, but we would like to point out that we com-

monly use 3-incision-procedures for VPS in our institution, with the cuttings at head, neck and abdomen. In some unpublished studies, this technique could lower the risk of tube occlusion and migration due to more convenient and safer intraoperative approaches, yet we still need more data to say whether it can reduce DICH in VPS patients or not.

A DICH after VPS is surely not an accident but a complication^{15,20}. Though the procedure is not a tricky one, hydrocephalus is an extremely complicated disorder itself and the same goes for DICH. Mechanisms of DICH still await exploring, confirmative conclusions are expected to be ascertained, whereas we doubt it will appear soon for the exceeding complexities and scarce nature of it.

Conclusions

DICH is a very rare complication of VPS procedure, which is potentially but not usually life-threatening. In our cases, the gradual erosion theory might be applicable for the explanation of Case 1, and accessorial brain fragility after trauma possibly accounts for the hemorrhage in Case 2. We recommend an onset longer than 48 hours necessary for a DICH diagnosis. Ultimate explanations for DICH following VPS are still in dispute. Treatment of this complication should follow general principles in neurosurgical care, meanwhile, we suggest routine CT scans on the same or the next day of VPS surgery, as well as regularly every 2 or 3 days after that. A preoperative MRI or cerebral angiography should also be considered if necessary.

Conflict of Interest

The Authors declare that they have no conflict of interests.

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