

## Rare Complications of VP Shunt Surgery Resulting in Significant Morbidity and Mortality: Report of Four Cases and Review of the Literature

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### Abstract

Ventriculoperitoneal Shunt surgery is a common neurosurgical procedure comprises a long list of postoperative complications. Among them, common complications like shunt infection, obstruction, overdrainage or underdrainage, subdural hematoma or hygromas are reviewed extensively in the previous literature.<sup>1</sup> With the course of time, improved surgical and technical skill, use of programmable valves, antibiotic impregnated catheters, attempt to improve nutritional status of the child before placement of shunt reduces significant shunt related morbidities. Despite all these precautions and techniques there are some rare complications of this surgery which can leads to catastrophic outcome. In this case series we reported four rare complications of VP shunt surgery and reviewed the previously reported literature. We also focused on the possible risk factors, pathophysiology of each complications so that meticulous attention can be given to prevent these complications to improve surgical outcome.

**Keywords:** VP shunt; Calcified extradural hematoma; Intracerebral hemorrhage; Ascities, Pseudocyst, SEGA

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### Introduction

VP Shunt Surgery is one of the most commonly performed neurosurgical procedure. Common

complications of VP shunt surgery are also reviewed in the literature like infection, migration, overdrainage, underdrainage, obstruction, etc.<sup>1-2</sup> Complications such as delayed intracerebral and intraventricular hemorrhage, extradural hematoma, shunt ascities, extraperitoneal pseudocyst are extremely rare but these complications can lead to severe morbidity and mortality of patient. Preoperative assessment, meticulous surgical technique, postoperative close monitoring and regular followup is necessary to minimize the complications. In our case series we have reported four rare complications of shunt surgery, discuss the possible risk factors and pathogenesis of each complications, reviewed the relevant literature so that we can set a standard management policy to optimize surgical outcome of this commonly performed neurosurgical procedure.

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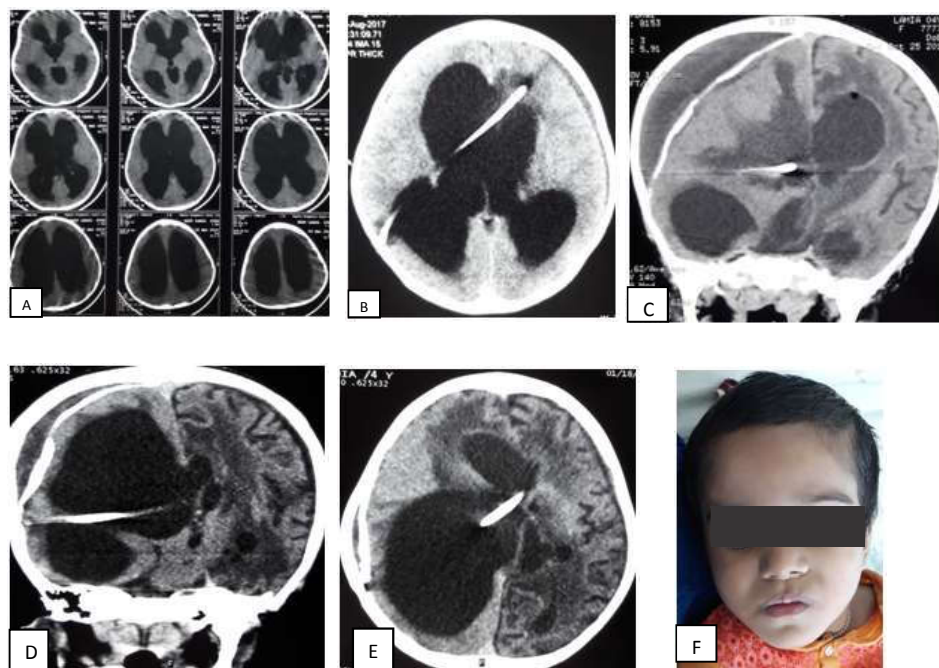
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## Case Series

### CASE 1

A 3-year-old girl admitted with sudden deterioration of level of consciousness for 5 days and fever for the same duration. She underwent right sided VP shunt surgery 1-year back due to obstructive hydrocephalus outside BSMMU. Neurological examination revealed: GCS-E1V1M3. All jerks were exaggerated and plantar response was bilaterally extensor. CT scan of brain showed calcified extradural hematoma with

asymmetric obstructive hydrocephalus (Fig. 1C). She underwent multiple parietal burrhole and drainage of liquefied hematoma. Postoperative course was uneventful. After 3 months, repeat CT scan of brain showed diminished size of the hematoma with improvement of sulcal and gyral pattern (Fig. 1D). During this time she underwent repeat VP shunt surgery in the same side. Patient's consciousness level was gradually improved and at 6 months follow-up GCS was E4V2M4. Still patient is under regular follow-up for further evaluation and management.

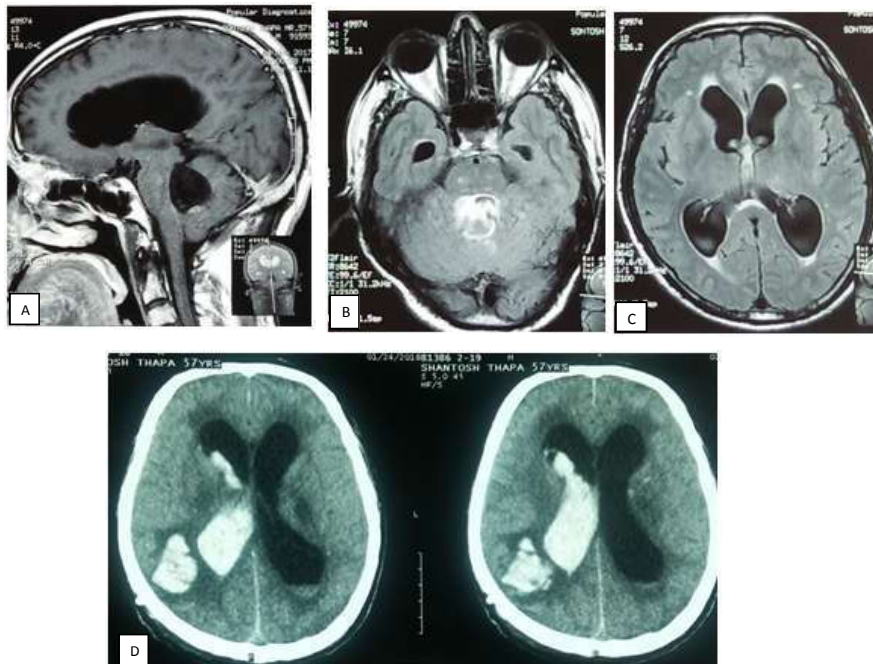


**Figs. 1A:** Obstructive Hydrocephalus, before placement of VP shunt; **1B:** Immediate postoperative CT scan of brain showing ventricular end in situ and mild diminution of the size of the ventricle; **1C:** CT scan of brain showing chronic calcified extradural hematoma, 1 year after placement of shunt with asymmetric obstructive hydrocephalus; **1D,E:** CT scan of brain showing diminished size of the hematoma and improvement of contralateral sulcal and gyral pattern at 3 months follow up; **1F:** Improvement of the consciousness level of the child.

### CASE 2

A 57-year-old gentleman, known case of hypertension and diabetes got admitted with the complaints of headache and balance difficulty for 2 months. He had positive signs of cerebellar dysfunctions and bilateral extensor plantar response. MRI of brain with contrast showed 4<sup>th</sup> ventricular space occupying lesion with features of obstructive hydrocephalus (Figs. 2A,B,C). Patient underwent posterior midline suboccipital craniectomy and gross total removal of tumor. 3 days after the definitive surgery, he underwent

right sided VP shunt surgery. Initially patient was symptomatically improved. His headache subsided and balance difficulty corrected. From 7<sup>th</sup> POD, patients consciousness level was gradually deteriorated. Urgent CT scan of brain showed intracerebral hemorrhage along the shunt tract with intraventricular hemorrhage (Fig. 2D). After resuscitation, patient was immediately transferred to operation theater and an EVD was placed into frontal horn through right sided Kocher's point. However, patient's condition was not improved and he expired on 9<sup>th</sup> POD.

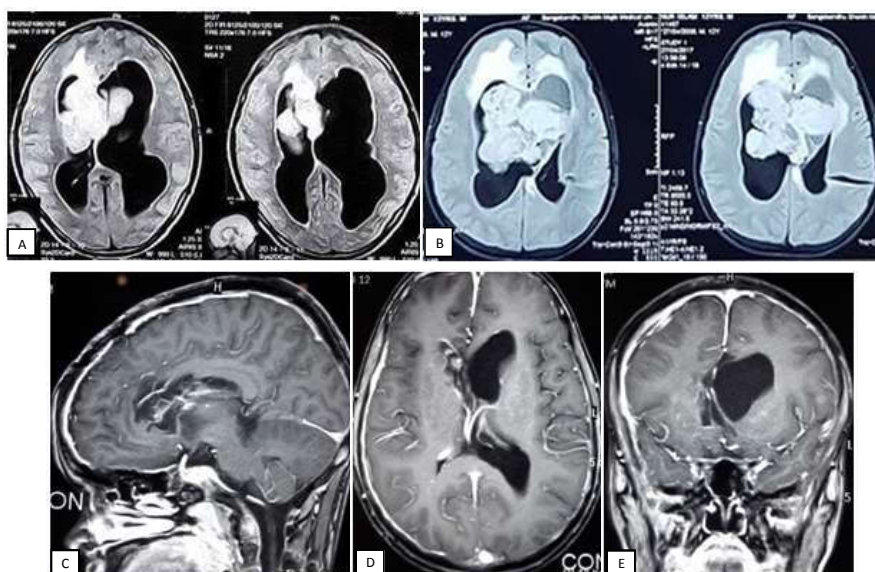


**Figs. 2A,B,C:** Preoperative MRI of brain with contrast showing 4<sup>th</sup> ventricular space occupying lesion with obstructive hydrocephalus; **2D:** Delayed intracerebral and intraventricular hemorrhage at 7<sup>th</sup> postoperative day.

**CASE 3**

A 12-year-old male patient presented with insidious onset of dull aching headache and vomiting for one year, progressive dimness of vision in both eyes for the same duration. He experienced one episode of generalized tonic clonic convulsion followed by loss of consciousness. After evaluation with MRI

of brain, he diagnosed as a case of SEGA with obstructive hydrocephalus and underwent urgent VP shunt surgery in a private hospital. However, 7 months following shunt surgery, patient admitted for definitive surgery into BSMMU with additional features of ascites. After admission, thorough examination showed visual acuity restricted to hand movement in both eyes and fundoscopy was



**Figs. 3A:** SEGA with obstructive hydrocephalus; **3B:** Diminution of the size of the ipsilateral ventricle after placement of VP shunt; **3C,D,E:** Follow-up MRI of brain after 1 year of definitive surgery showed no tumor recurrence and well functioning VA shunt.

consistent with features of secondary optic atrophy. Per abdominal examination revealed shifting dullness and positive fluid thrill. However, his liver function test, renal function test and cardiac evaluation were normal. Serum albumin, total protein and albumin-globulin ratio was also within normal limit. At this stage, we exteriorize the lower end of VP shunt and drain 1 litre ascitic fluid for decompression. We sent both CSF and ascitic fluid for biochemical, cytological and microbiological examination. Findings were normal except high protein level both in CSF and ascitic fluid. Later on, we came into conclusion that, CSF high protein level was responsible for impaired peritoneal absorption of CSF. Patient underwent anterior interhemispheric transcallosal approach and gross total removal of tumor. At 10<sup>th</sup> POD, he underwent right sided ventriculo-atrial shunt. His postoperative period was uneventful. 1 year after surgery, repeat MRI of brain with contrast showed no recurrence of tumor with well functioning status of the shunt (Figs. 3 C,D,E)

#### CASE 4:

A 5-month-old boy presented with the features of repeated vomiting since birth and diagnosed as a case of congenital obstructive hydrocephalus. He underwent right sided VP shunt surgery at the age of 15 days. After 3 months, revision of VP shunt done due to shunt obstruction. His postoperative period was uneventful and he was symptomatically improved for next 2 months. Then he developed a small reddish swelling along the shunt tract in the anterior abdominal wall (Fig. 4C). Baby was symptomatically deteriorated and repeat CT scan of brain showed ventricular size was enlarged than before. Baby underwent re-exploration of the lower end of VP shunt. There was a pseudocyst present beneath the visible swelling which was responsible for shunt malfunction. Lower end was exteriorized and after 1 month, left sided VP shunt surgery done. After 3 months follow-up, shunt was functioning well.

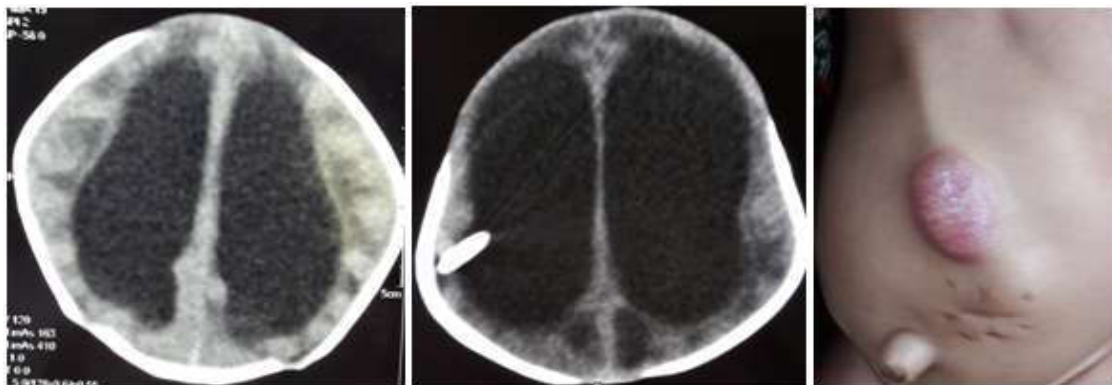


Fig. 4A: Obstructive hydrocephalus, before placement of VP shunt; 4B: Hydrocephalus became more prominent 6 months after placement of the shunt; 4C: Extraperitoneal anterior abdominal wall pseudocyst.

#### Discussion

Although much attention paid regarding common complications of VP shunt surgery in the previously reported literature-like infection, migration, overdrainage, underdrainage, obstruction, etc.,<sup>1,2</sup> least attention given towards the rare complications, like- extradural hematoma, intracerebral hemorrhage, intraventricular hemorrhage, tumor induced hyperproteinorrhachia and extraperitoneal pseudocyst. Only some isolated case reports are available regarding each complications but no large studies were done which could effectively explain the possible aetiological factors and management

protocol. Recently, one large study was conducted by Gong *et al.* regarding intracerebral hemorrhage after placement of VP shunt in which he discussed about the risk factors and some surgical strategied to overcome this catastrophic complications.<sup>18</sup>

Although subdural hematoma is much commoner than epidural hematoma after VP shunt surgery due to rapid decompression of intracranial compartment, despite this epidural hematoma may form because of loose adhesion of dura with the inner surface of skull in some patient.<sup>1,3</sup> A sudden lowering of intracranial pressure leads to cortical collapse which helps expansion of hematoma increasing up to a catastrophic complication if not

recognized and treated in time. In the previously reported literature, younger age and presence of chronic hydrocephalus was considered to be the possible risk factors because at younger age, dura become loosely adhere with the skull.<sup>3,7</sup> Reported incidence of epidural bleeding is 5.6% in the series of Driesen and Elies.<sup>8</sup> Though use of programmable shunt valve significantly lower the incidence of epidural hematoma and previously, 18 reported cases of epidural hematoma were found despite using programmable valve.<sup>3,4,8-15</sup> Most of the cases were acute extradural hematoma and among them, only 6 cases were chronic ossified extradural hematoma including our own (Table 1).

In our case possible etiology could not be evaluated well as she underwent VP shunt surgery outside BSMMU. Despite this, age could be a possible factor. However, there are several proposed techniques to prevent this complication. Driesen and Elies<sup>8</sup> suggested to fix the dura adjacent to the burr hole to minimize bleeding complication. Other precautions suggested by the Kalia *et al.* that: minimal CSF spillage at the time of ventricular catheter insertion; meticulous surgical technique; use of high or medium pressure valves; slow return to the full upright position and a close follow-up with postoperative CT-scan.<sup>3,6</sup>

**Table 1:** Reported Cases of Chronic Ossified Extradural Hematoma after Placement of VP Shunt

Authors	Year of publication	Age (yrs)	Sex	Site of VP shunt placement/other causes	Time interval for detection	Site of ossified extradural hematoma	Outcome
Mathuriya <i>et al.</i> <sup>4</sup>	1989	35	M	Right sided parietal	3 months	Right sided parietal	Satisfactory
		29	M	Right sided parietal	5 weeks	Right sided parietal	Satisfactory
Kalia <i>et al.</i> <sup>3</sup>	1993	15	F	Right frontal ventriculoperitoneal shunt with a medium pressure (104 mm H <sub>2</sub> O) Hakim valve	3 months	Bifrontal	Complete resolution of symptoms
Pereira <i>et al.</i> <sup>6</sup>	1998	33	F	Left sided ventriculoperitoneal shunt with medium pressure valve	4 months	Left parietal	Patient denied further management. Symptomatic treatment given
Seyithanoglu <i>et al.</i> <sup>5</sup>	2010	17	F	Right temporo-occipital ventriculoperitoneal shunt with a medium pressure flow control valve (Medtronic)	3 years	Bifrontal	Satisfactory
Barua KK	2018	3	F	Right sided keen's point with medium pressure Chabra VP shunt	1 year	Right parietal	Satisfactory

Intraventricular hemorrhage following removal of VP shunt is well documented in the previously reported literature.<sup>17</sup> However, Gong *et al.* first conducted the largest study regarding characteristics of delayed intracerebral hematoma following placement of VP shunt. Among 754 patient, 12 patient developed DICH and reported incidence in his series is 1.59%.<sup>18</sup> Matsumura reported the first case in 1985.<sup>22</sup> From 1985 to 2017 we have reviewed the reported cases of DICH who developed hematoma at or after 5 days of VP shunt placement (Table 2). Possible etiological factors related to age, history of craniotomy and manipulation of the valve system were proposed. However, the suggested mechanism is that after craniotomy operation and following VP shunt placement, there is erosion of

cerebral vessels following collapse of the ventricles which is responsible for the development of DICH, specially if the age of the patient is above 60 years.<sup>18</sup> The DICH is an uncommon complication of VP shunt placement. Gong reported only 8.3% mortality in his series<sup>18</sup> but percentages were more is the previously reported cases. The suggested risk factors of acute intracerebral hemorrhage after VP shunt placement are coexistent bleeding disorder, shunt induced disseminated intravascular coagulation, disruption of an intracerebral tumor, disruption of intracerebral vessel, hemorrhage from an occult vascular malformation and head trauma occurring shortly after shunt placement.<sup>29-31</sup> However, these risk factors failed to explain the delayed hemorrhage. According to previous

**Table 2:** Reported Cases of Delayed ICH/IVH (Defined as IVH  $\geq$  5 days) after VPS Placement

Authors	Year of publication	Age (years)	Sex	Onset of hemorrhage	IVH and/or ICH
Matusmura <i>et al.</i> <sup>22</sup>	1985	17	Male	7	IVH and ICH
		43	Female	5	IVH and ICH
Snow <i>et al.</i> <sup>23</sup>	1986	68	Male	15	IVH and ICH
		64	Female	6	IVH and ICH
Mascalchi <i>et al.</i>	1991	64	Male	14	IVH and ICH
Alc'azar <i>et al.</i> <sup>20</sup>	2007	0	–	22	IVH and ICH
		32	Female	5	IVH and ICH
Misaki <i>et al.</i> <sup>24</sup>	2010	0	–	7	IVH and ICH
Khandelwal <i>et al.</i>	2011	69	Male	8	IVH
Zhou <i>et al.</i> <sup>21</sup>	2012	78	Male	4	IVH
Okazaki <i>et al.</i>	2013	76	Male	7	ICH
Ma <i>et al.</i>	2015	67	Male	5	ICH
Mavridis <i>et al.</i>	2017	65	Male	7	ICH
		68	Female	5	ICH
Gong <i>et al.</i> <sup>18</sup>	2017	61	Male	5	ICH

studies, some results suggested that the fragility of brain and the erosion of a cerebral blood vessel might be involved in the risk factors.<sup>21,24</sup> In our case, age and history of craniectomy would be the possible etiological factor.

The mechanical complications results as a consequence of disconnection, breakage or migration of the system. Distal catheter of the VP shunt can give rise to abdominal abscesses, perforation of the hollow of viscus, intestinal obstruction, peritonitis, ascites, and peritoneal CSF pseudocyst formation.<sup>28,29</sup> In addition, the migration of peritoneal catheters are reported through large intestine, anus, vagina, umbilicus and inferior vena cava.<sup>30</sup> Intraperitoneal CSF pseudocysts may develop at the distal end of the catheter from a few weeks to a year after the VP shunts are inseted.<sup>30</sup> The clinical condition usually begins with diffuse abdominal pain and continues with the development of neurological symptoms specially with features of raised intracranial pressure. Laparoscopic excision of intraperitoneal cysts followed by relocation of the catheter are considered best treatment for CSF peritoneal pseudocysts. In the brief review of the relevant literature, anterior abdominal wall extraperitoneal pseudocyst is extremely rare. Lee first reported a large extraperitoneal pseudocyst in 2015, which was successfully treated by lower end revision with cyst excision and aspiration. In his reported case, the distal end of the catheter was exceedingly short, and thus the catheter easily migrated from the peritoneal cavity. The function of the VP shunt was patent and the large CSF pseudocyst appeared to develop under the relatively low pressure of

the anterior extraperitoneal space, compared with the intracranial pressure, and the CSF dissected through the extraperitoneal fascia. Therefore, the large pseudocyst developed gradually.<sup>31</sup> Compared to this, in our reported case there was features of underdrainage of CSF because of superimposed infection. So, lower end was exteriorized, control of infection done followed by revision of shunt done in healthy site.

Hydrocephalus is the main factor responsible for clinical symptoms in symptomatic SEGA patient rather than the tumor volume itself. However, over time, the approach to these lesions has become changed to initial gross total resection which can avoid the placement of VP shunt. Recent studies showed some correlation between SEGA and VP shunt malfunction due to overproduction of protein which is responsible for impaired peritoneal absorption of CSF.<sup>32-34</sup> Perek Polnik *et al.* first reported a case of SEGA who had an external CSF drainage for over 6 months due to sustained increased CSF protein level of 1200 mg/dL. Later on, the boy treated with everolimus therapy which significantly reduced tumor volume, thus decrease the level of CSF protein concentration.<sup>33</sup> Thus, an abnormally high level of CSF protein in nonresected SEGA leads to the frequent obstruction of CSF diversion devices in shunted patients as well as impaired peritoneal absorption giving rise to frequent revision of VP shunt if not properly evaluated. Laviv *et al.* also reported 2 cases of SEGA who underwent VP shunt revision surgery for multiple times. Later on, on searching the possible aetiological factors, it was discovered that high CSF protein level was responsible for the

device failure. Surgical decompression followed by everolimus therapy achieved significant control on tumor volume, thus associated with spontaneous reduction of CSF protein level. Based upon the previous literature, in our reported case,

we went for ventriculoatrial shunt and no valve was placed. After one year follow-up, there was no recurrence of tumor and well functioning of the VA shunt. There was no features of endocarditis as well.

**Table 3:** Reported Cases of CSF Ascities due to Hyperproteinorrhachia Caused by SEGA

Authors	Year of publication	Age	Sex	Management	Outcome
Perek Polnik M <i>et al.</i> <sup>33</sup>	2012	10	Male	External CSF drainage followed by treatment with everolimus	Satisfactory
Laviv Y <i>et al.</i> <sup>32</sup>	2015	21	Male	Anterior interhemispheric transcallosal approach and subtotal resection for 2 times, VP shunt revision for 2 times followed by everolimus therapy	Satisfactory
		24	Female	Anterior interhemispheric transcallosal approach and gross total resection, VP shunt revision for 2 times followed by everolimus therapy	Satisfactory
Kasper E <i>et al.</i> <sup>34</sup>	2017	18	Female	Shunt revision followed by right frontal craniotomy and removal of tumor through transcortical transventricular approach	Satisfactory
Barua KK <i>et al.</i>	2018	12	Male	VP shunt followed by anterior interhemispheric transcallosal approach and gross total removal of tumor. VA shunt done due to VP shunt malfunction	Satisfactory

**Conclusion**

From our experience, we have tried to focus on the rare complications of this commonly performed neurosurgical procedure which can lead to catastrophic outcome if timely intervention is not taken. We have also reviewed the previously reported cases and discuss the possible risk factors of each complications but larger studies are required to standerzize the management protocol.

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**Abbreviation**

- BSMMU Bangabandhu Sheikh Mujib Medical University
- CT Computed Tomography
- CSF Cerebrospinal fluid
- DICH Delayed Intracerebral Hemorrhage
- MRI Magnetic Resonance Imaging
- POD Postoperative Day
- SEGA Subependymal Giant Cell Astrocytoma
- T1WI T1 Weighted Image
- T2WI T2 Weighted Image
- VA Ventriculo atrial
- VP Ventriculoperitoneal

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