



Case Series

Shunt exposure as a ventriculoperitoneal shunt complication: A case series



Wihasto Suryaningtyas, I.G.M. Aswin R. Ranuh, Muhammad Arifin Parenrengi*

Department of Neurosurgery, Faculty of Medicine, Universitas Airlangga, Dr Soetomo Academic Medical Center Hospital, Indonesia

ARTICLE INFO

Article history:

Received 16 December 2020

Received in revised form 20 January 2021

Accepted 20 January 2021

Available online 27 January 2021

Keywords:

Exposed shunt

Ventriculoperitoneal shunt

Hydrocephalus

Case report

ABSTRACT

INTRODUCTION: Shunting is a technique in neurosurgery for treating hydrocephalus. Shunting is an effective choice for both cases of obstructive or communicative hydrocephalus. However, in some rare cases, complications, such as exposed shunt, can occur. In this case series, the author discusses 6 cases of hydrocephalus patients with exposed shunts. The risk factors, diagnosis, and management of exposed VP shunt will be discussed further in this case series report.

METHODS: This study was an analysis of all cases treated in a period of 1 year from January to December 2018 with an inclusion criterion of history of exposed shunt of any age group. This study is a single-centre retrospective report of the clinical presentation and radiology examination before and after treatment. Clinical and radiology evaluation were performed in immediate post procedural period. A detailed clinical examination was performed to look for exposed shunt complication.

RESULTS: In this series of studies 6 patients with hydrocephalus who had shunts were reported. All patients were pediatric patients. Six patients had exposed shunt on the scalp or abdomen. Radiological examinations including CT scan, chest X-ray or babygram were performed to evaluate the location of the shunt. The data reported was from January to December 2018 and there were 301 VP shunt installment cases. Management included surgical revisions and treatments to prevent further complications.

CONCLUSION: Shunting is still a routine therapy in the field of neurosurgery, although other modalities such as endoscopic third ventriculostomy (ETV) have started to be performed more frequently. Complications such as exposed shunt are rare in the treatment of pediatrics with hydrocephalus. We presented that exposed shunt is a rare complication (2.3% incidence rate) which might be caused by certain risk factors, such as age of patient when the shunting was performed, and nutritional status. Early diagnosis and treatment are important to prevent further complications, especially infections. Subpericranial technique for shunt tunneling might be useful in preventing exposure of shunts with associated morbidity factors.

© 2021 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Background

1.1. Introduction

Hydrocephalus is the accumulation of cerebrospinal fluid (CSF) in the brain's ventricular system. An increase in the amount of excess CSF will result in increased intracranial pressure (ICP) and also suppression of the surrounding normal tissue [1]. The cause could be in production disruptions or flow disturbances or return absorption. Hydrocephalus can be congenital or acquired; generally associated with other diseases so the cause must be sought. Hydrocephalus, based on its CSF flow patency, is divided

into two types; non-communicating or obstructive hydrocephalus. The etiology of hydrocephalus includes congenital etiology or acquired. Congenital causes include neural tube defects, arachnoid cysts, Dandy-Walker syndrome, and Arnold-Chiari malformations. The etiology of acquired hydrocephalus include brain tumors, meningitis, brain abscess, head injury, and non-traumatic intracranial bleeding. Another type of hydrocephalus is normal pressure hydrocephalus (NPH), entrapped fourth ventricle and arrested hydrocephalus [2].

Hydrocephalus can occur in all age groups, generally in children [1]. Symptoms of hydrocephalus may be different in each age group. In the children age group, the most visible symptom is enlarged head circumference. The clinical presentation of pediatric hydrocephalus varies depending on each individual cases. The clinical presentations include abnormalities in head/occipital frontal circumference (OFC), enlarged cranium at rate > facial growth, irri-

* Corresponding author.

E-mail addresses: aswinranuh@rocketmail.com (I.G.M.A.R. Ranuh), arifin_ns@yahoo.com (M.A. Parenrengi).

tability, poor head control, full and bulging fontanelle and sun sign setting (upward gaze palsy) [2].

Neurosurgical management of hydrocephalus includes shunt and ETV (endoscopic third ventriculostomy). Ventriculoperitoneal (VP) shunt is the most common surgical procedure performed for hydrocephalus cases. Shunting or drainage of CSF into another cavity is one of neurosurgical techniques for the treatment of hydrocephalus, is the choice of surgery effective both for cases of obstructive or communicating hydrocephalus. ETV (endoscopic third ventriculostomy) is another option besides shunt. ETV is a procedure to make a hole in the third ventricular floor using an endoscope placed inside the ventricular system through a burr hole, so that CSF flows directly into the basal cisterns through the blockage. ETV is the main choice for cases of non-communicating hydrocephalus [4–7].

The most common complication of VP Shunt is shunt malfunction, in which the device fails to serve its purpose and, should it not be revised, would leave the patient deteriorate. VP Shunt failure could be due to obstruction or infection. There are many factors which could contribute to the development of shunt infection, one of which is shunt exposure through the scalp [7].

Although shunt exposure itself is a risk factor of shunt infection, it could occur the other way around. An existing infection around the shunt material may give way to shunt exposure. Continuous stress on the skin, especially at a region where the skin is thinner and more fragile, will eventually lead to shunt exposure [1,4,5].

Although shunt exposure is relatively rare, its management could be difficult. If left untreated, shunt exposure might cause scalp necrosis or suture dehiscence [3–5]. This case-series aims to illustrate our experience in dealing with shunt exposure in Dr. Soetomo Academic Medical Center Hospital, Surabaya.

1.2. Literature review

1.2.1. Anatomy

The ventricular system is a set of cavities where cerebrospinal fluid (CSF) is produced and circulate within the brain. Within the ventricular system, a group of specialized ependymal cells known as choroid plexus is responsible to produce the CSF. This fluid gives mechanical protection, regulates homeostasis within the cerebral interstitial fluid, and facilitates brain development. Cerebrospinal fluid travels through the ventricular system in a unidirectional, rostrocaudal fashion and ultimately communicates with the cranial and spinal subarachnoid space, which is the predominant location of CSF reabsorption [6].

The cerebral ventricular system is made up of 4 ventricles: (1) Two lateral ventricles (1 in each cerebral hemisphere), (2) the third ventricle in the diencephalon, and (3) the fourth ventricle in the hindbrain. Inferiorly, it is continuous with the central canal of the spinal cord [6].

The paired lateral ventricles consist of an anterior, inferior, and posterior horns. The three horns give the ventricles a C-shape. Each horn extends peripherally in the frontal, temporal, and occipital lobes, respectively. The lateral ventricles communicate with the third ventricle through the foramen of Monroe. The anterior, or frontal, horns of the lateral ventricle extend from the frontal lobe, connecting with the body of the lateral ventricle at the level of the foramen of Monroe [6]. The shape and size of the foramina correlate with the size of the ventricles: If the ventricles are small, each foramen will be a crescent-shaped opening anteriorly bounded by the concave curve of the fornix and posteriorly by the convex anterior tubercle of the thalamus. As the ventricles enlarge, the foramen on each side becomes more rounded. Structures passing through the foramen include the choroid plexus, the distal branches of the medial posterior choroidal arteries, and the, thalamostriate, superior choroidal and septal veins [7].

The corpus callosum surrounds the frontal horn as the superior, anterior, and inferior boundaries. The corpus callosum consists of the genu, body, and splenium, and a small portion called the rostrum which extends inferiorly and posteriorly from the genu. The genu delineates the frontal horns of the lateral ventricles superiorly and anteriorly, while the rostrum inferiorly. Between the frontal horns lie the septum pellucidum. Lateral to the frontal horns is the septum pellucidum [7].

The bodies of the lateral ventricles are located superior to the thalamus. The atrium of the lateral ventricle extends from the posterior aspect of the body and is contiguous with the paired inferior (temporal) and posterior (occipital) horns. The inferior (temporal) horns extend inferiorly from the atrium, wrapping around the pulvinar nuclei of the thalamus before terminating in the anterior temporal lobes [8].

The cerebral aqueduct of Sylvius connects the third ventricle to the fourth ventricle. Due to its narrow nature, obstruction of the aqueduct of Sylvius would easily causes hydrocephalus. The cerebral aqueduct traverses the dorsal aspect of the midbrain, surrounded by the periaqueductal gray matter, which is situated anterior to the tectum and posterior to the tegmentum [6,8].

The fourth ventricle is a tent-like cavity of the hindbrain filled with CSF. It is posterior to the pons and cranial half of medulla and anterior to the cerebellum. On a sagittal section, it appears like a triangle, while on a horizontal section it mimics a diamond. It receives CSF from the third ventricle through the aqueduct, and is continuous with the central canal of the spinal cord. It's drained into the subarachnoid space by the paired lateral foramina of Luschka and the single midline foramen of Magendie.

The fourth ventricle is neighbored inferolaterally by gracile and cuneate tubercles and inferior cerebellar peduncles and superolaterally by the superior cerebellar peduncle. The cephalic portion of the roof is formed by 2 superior cerebellar peduncles whose medial margins overlap the ventricle on reaching the inferior colliculi. The superior medullary velum bridges the gap between superior cerebellar peduncle [7]. The lower half of the roof is covered by a thin sheet of non-nervous tissue, the inferior medullary velum, where the tela choroidea of the fourth ventricle resides. Laterally, it meets the inferolateral border of the ventricular floor marked by a white ridge called Taenia. Inferiorly, the two taeniae meet to form a small fold called obex while superiorly it passes along the lateral recess. The plexus assumes a T-shape with a vertical and horizontal limb. The horizontal limb continues in the lateral recess through the aperture foramen of Luschka into the subarachnoid space. In certain cases, this foramen is closed by a membrane. This forms a pouch containing choroid plexus at the cerebellopontine angle called Bochdalek's flower basket. Enlargement of this pouch may give rise to clinical symptoms [8].

1.2.2. Epidemiology of VP shunt complication and etiology

Hamdan et al. reported cases with exposure of skin overlying the tube occurring in 3 of 205 patients with VP shunts [5]. VP shunt exposure is more common in children than adults. Shunt exposure on scalp is one of the scalp complications and this is rare. Complications that can be caused by shunt exposure are necrosis, suture dehiscence and infection. There are several things that cause VP shunt exposure. This is attributed to the thinner skin of the child's head so that it is easier for the tube to protrude. In children with hydrocephalus, the scalp becomes thinner so that VPS exposure becomes more likely [6].

Shunt complications on pediatrics has 4.22-fold greater odds of shunt revision compared to adults. The most common complications of shunt placement in both pediatric and adult are shunt malfunction caused by obstruction. The second most common cause of shunt malfunction is infection. Infection is the second common cause of shunt malfunction, which ranges from 8 to 15% among

patients who undergo VPS placement. Shunt malfunction is most often caused by obstruction [4,5,10]. Obstruction can occur due to over-drainage which causes debris from being pulled into the proximal catheter. Currently developing programmable shunts that can reduce this risk. In addition to the proximal tube, occlusion of the distal tube can also occur due to accumulation of debris obstruction, such as blood and proteinaceous fluid [5–7,10]. Several factors are related to the occurrence of VP exposure. One factor is infection. Infection can occur as a risk factor for VP shunt exposure and infection can also occur as a complication of VP shunt exposure. Infection causes changes in the function of the skin as a protector. The manifestation of an infection is an abscess and skin fistula which if caused a VP shunt exposure. Continuous pressure on the skin can also contribute. Pressure will cause interference with blood flow and ischemia. Ischemia causes the skin to become more fragile and thinner which eventually will be necrosis [8].

1.2.3. Pathophysiology of hydrocephalus

In the most familiar model of cerebrospinal fluid (CSF) flow, CSF is produced by choroidal plexus, located within the lateral, third, and fourth ventricles. CSF travels slowly and unidirectionally through the ventricular system, exits the fourth ventricle into the subarachnoid space, and is absorbed through arachnoid granulations into the venous sinuses and systemic circulation. In this model, hydrocephalus is a consequence of physical or functional obstruction within the ventricular system, the subarachnoid space, or the venous sinuses [9]. Within the ventricular system, an obstructive malformation or gliosis can cause physical blockage of CSF flow. Outside the ventricular system, inflammation and scarring of the subarachnoid space, or elevated pressures within the venous sinuses, can impair translocation of CSF into the systemic circulation, leading to be hydrocephalus [10].

Especially, up to one third of CSF exits the skull along cranial nerve sheaths and into the lymphatic system rather than into the venous sinuses. Disturbances of this lymphatic exit pathway have been implicated in rat models of hydrocephalus, but whether they play a role in the pathogenesis of human hydrocephalus is not yet known. Superimposed upon unidirectional CSF flow is pulsatile movement of CSF during the cardiac cycle [9,10]. With each beat of the heart, CSF flows through the foramen magnum, into the spinal subarachnoid space and then back into the skull again. CSF has also been found to flow in a pulsatile manner from the intracranial subarachnoid space into the brain parenchyma, and then into the systemic circulation, along perivascular pathways. Augmentation of pulsatile ventricular pressure waves in animal models results in hydrocephalus. Altered pulse of CSF flow has repeatedly been described in conjunction with human hydrocephalus, but whether it is cause or consequence remains unclear [9–11].

1.2.4. Clinical manifestation and radiology

Clinical manifestation may vary with age. Fetal ventriculomegaly can be identified by prenatal ultrasound as early as 18–20 weeks' gestation. Detection also followed by another examinations, including a level two ultrasound scan, fetal MRI, TORCH (toxoplasmosis, rubella, cytomegalovirus, herpes simplex) screening, or amniocentesis. In known maternal carriers of L1CAM mutation, chorionic villus sampling or amniocentesis can be offered for prenatal diagnosis of X-linked hydrocephalus [12]. In infants, signs and symptoms of hydrocephalus presents with an abnormally increasing head circumference, irritability, vomiting, bulging of the anterior fontanel, or splaying of the cranial sutures. True hydrocephalus must be distinguished from so-called benign external hydrocephalus or benign enlargement of subarachnoid space, which needs no treatment and is characterized by enlarged subarachnoid spaces, only mild or absent ventriculomegaly, and a clinically well child [9,11]. Beyond infancy, hydrocephalus typi-

cally presents with a constellation of findings that include some combination signs like headache, vomiting, loss of developmental milestones, diplopia (usually from a VI cranial nerve palsy), or papilledema. Brain imaging is the most important diagnostic investigation. An infant with an open fontanel can be screened for ventriculomegaly by cranial ultrasonography, but an MRI study (MRI became gold standard for hydrocephalus case because avoids radiation exposure and provides more information) is typically indicated to elucidate the anatomy and cause. Cine MRI CSF flow imaging might provide insight into patient-specific changes in CSF hydrodynamics and, particularly in cases where a site of obstruction is questionable, these methods can inform surgical decision making and provide a means to assess treatment efficacy [12].

1.2.5. Treatment

Hydrocephalus could be treated medically and surgically. Medical treatment of hydrocephalus aims to reduce the production of CSF and dehydrate the brain. Various drugs have been tested for hydrocephalus, among which are isosorbide, glycerol, acetazolamide, and steroid [13,14].

Ventriculoperitoneal (VP) Shunt is the main treatment modality for hydrocephalus. It dramatically improves neurological outcome despite being associated with several complications [12]. VP shunt is utilized to divert CSF. It is basically a tube with a pressure-regulating valve that carries CSF from the ventricular system to another space outside the brain, such as the peritoneum [2]. Surgery is done with general anesthesia. The position of the patient is supine, with the head being rotated to the contralateral side. Used a carbon head clamp adapter and Mayfield carbon head clamp. Evaluation is based on a CT scan to check the ventricular catheter position [15]. For small children, the length of the peritoneal catheter is generally 30 cm length of intra-peritoneal tubing to allow continued growth (120 cm total length of peritoneal tubing was associated with a lower revision rate for growth without a significant increase in other complications) [16].

Indication of VP shunt are as follows; congenital hydrocephalus, tumors leading to CSF blockage of the lateral or third ventricles, the posterior fossa, and intraspinal tumors, post-hemorrhagic hydrocephalus, spina bifida causing hydrocephalus, congenital aqueductal stenosis, craniosynostosis, post-meningitic hydrocephalus, Dandy-Walker, arachnoid cysts, and Idiopathic intracranial hypertension [4,5,15].

2. Case reports

This study is an analysis of 6 cases of hydrocephalus treated with shunting, leading to a complication of exposed shunt. Statistical analysis was collected from all VP shunt installment in Dr. Soetomo Academic Medical Center Hospital. This study has been reported in line with the SCARE and PROCESS 2018 criteria [17]. All VP shunt procedures were performed by our senior neurosurgeons who already have undertaken the additional training in pediatric neurosurgery subspecialization. This study involves pediatric patients with VP shunt insertion prior to hospital admission. The data reported were from January to December 2018, and there were 301 VP shunt installment cases in our academic medical center, Dr. Soetomo hospital. Among those cases, there were 6 exposed shunt cases. All patients' caretakers consented to their data being taken & publicized. The incidence of exposed shunt was 1.9% (6/301). Elective VP shunt installment were 71 cases and emergency VP shunt installment were 230 cases. Assessed prevalence of each risk factor in this case report include the age of when the VP shunt was installed, nutritional status at birth, and hypoalbuminemia.

The first case was a 2-year-11-month female who had a diagnosis of hydrocephalus. The patient underwent VP shunting when

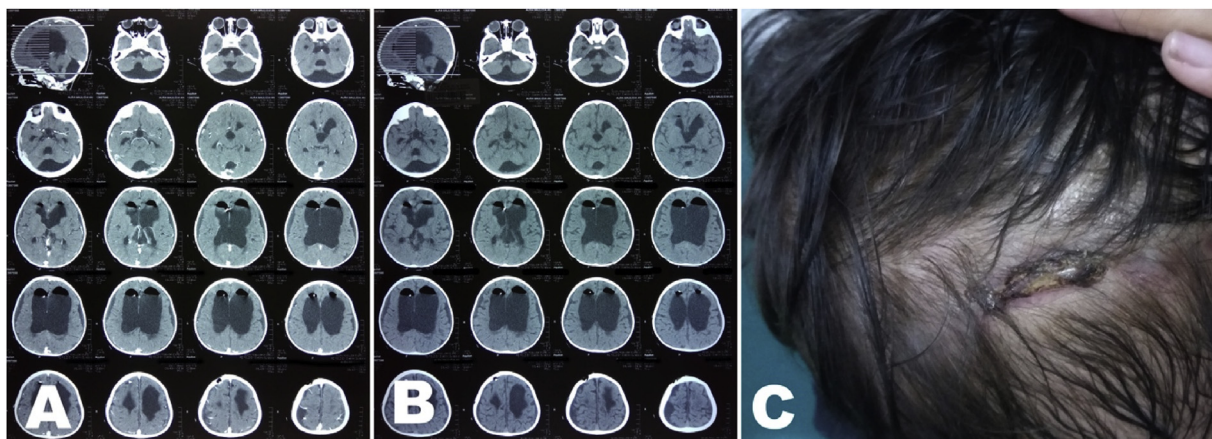


Fig. 1. The first case, a three-year-old female, who had a diagnosis of hydrocephalus treated with ventriculoatrial (VA) shunt and had an exposed shunt.; A and B Pre-procedural CT-Scan; C. Pre procedural clinical image showing tube protrusion on scalp.

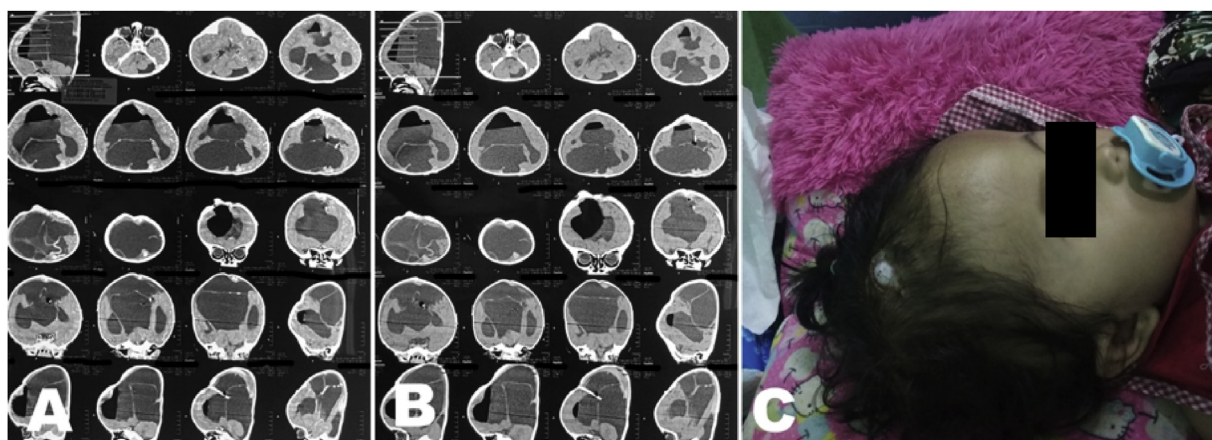


Fig. 2. The second case, 15-month-old girl baby comes with a diagnosis of hydrocephalus with VA shunt. The second case; A and B Pre procedural CT Scan; C. Pre procedural clinical image showing tube protrusion on scalp.

she was 8 months old. VP shunt was removed, and when she was 2 years old and it was changed to an EVD. EVD was removed 2 weeks afterwards and a ventriculoatrial (VA) shunt was installed. VA shunt was installed 8 months before admission. The patient came with a chief complaint of a wound on the scalp skin for the last 2 months ago prior to admission. The patient has already received local infection control such as routine wound cleansing by normal saline, and sterile gauze swab as needed. The shunt tube was visible from outside the scalp for the last 2 weeks. The patient also had an intermittent fever for the last 2 months. Weakness of the right extremities was complained for the past 2 years. Her body weight was 15 kg. Patient was planned to undergo VA shunt removal and VP shunt insertion. The patient had improved clinical outcome and was discharged from the hospital. The patient was followed up 6 months later with improved exposed site and a satisfactory result was reported from the parents (Fig. 1).

The second case was a 15-month-old girl baby who came with a diagnosis of hydrocephalus with VA shunt. Patients got seizures 1 week before admission. The patient was treated for 4 days in the hospital prior to VP shunt insertion. The patient had a VP shunt when she was 3 months old, and a VA shunting along with endoscopic fenestration was performed 10 months before admission. Her body weight was 9 kg. The patient was planned for VA shunt removal and external ventricular drain (EVD) installation. The patient had improved clinical outcome and was discharged from hospital care. The patient was followed up 6 months later

with improved exposed site and a satisfactory result was reported from the parents (Fig. 2).

The third case was a 2-month-old baby girl with a diagnosis of hydrocephalus and VP shunt, who came to the emergency room with a complaint of a reddish wound on the scalp 1 week ago. Her body weight was 5 kg. History of fever was found in the patient. Patient got seizures 12 h before admission. The patient had a history of VP shunt installation when she was 1 months old. The patient also had a history of lumps at the back of her head at birth and at the age of 8 days, she underwent cele excision. The patient was given the antipyretic agent and amount of fluids based on her body daily needed. Patient was planned & operated for shunt removal, debridement and reinsertion of VP shunt. The patient was followed up 6 months later with improved exposed site and a satisfactory result was reported from the parents (Fig. 3).

The fourth case was a 9-month-old baby boy, who came with a chief complaint of wound on insertion location on scalp. Initially only reddish spots appeared in the last 1 month before admission. Tube appeared from the scalp since 1 week ago. Her body weight was 8 kg. Patient had a history of surgery of subdural drainage and subdural peritoneal shunt installation when she was 2 months old. The patient had improved clinical outcome and was discharged from hospital care. The patient was followed up 3 months later with improved exposed site and a satisfactory result was reported from the parents (Fig. 4).

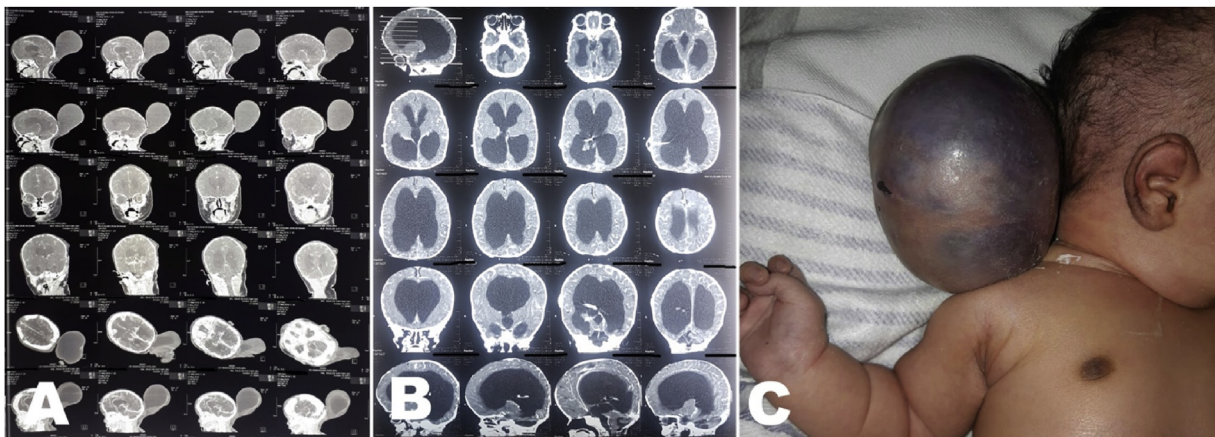


Fig. 3. The third case, 2-month-old baby girl with a diagnosis of hydrocephalus and cele and underwent VP insertion and cele excision. VP shunt came to emergency room with reddish wound on scalp a week ago. Patient planning for shunt removal, debridement and reinsertion of VP shunt; A and B Pre and post procedural CT Scan of VP shunt insertion and cele excision; C. Pre procedural clinical image.

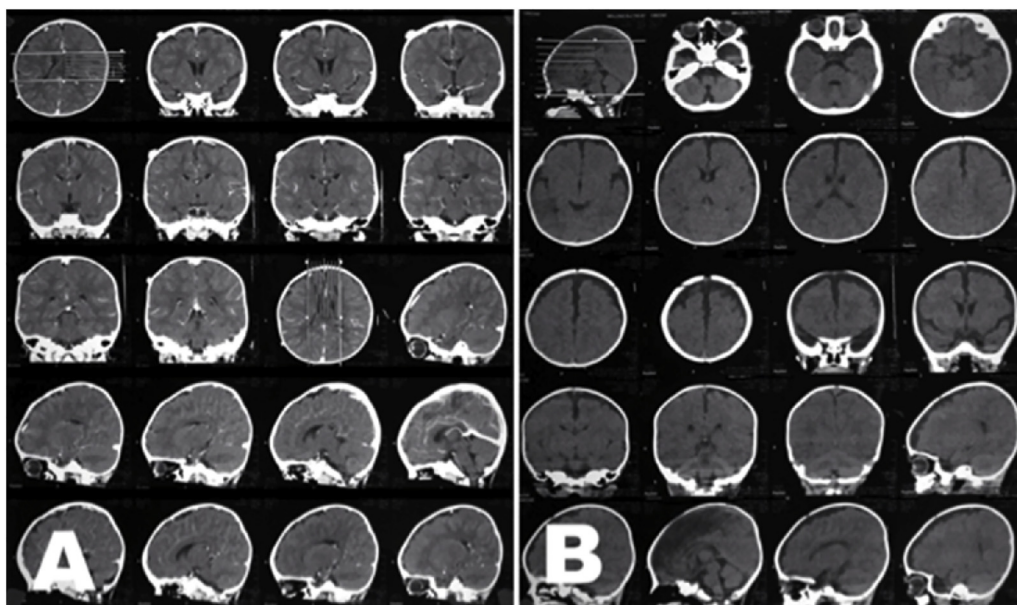


Fig. 4. The fourth case, 9-month-old baby girl with a diagnosis of hydrocephalus and VP shunt came to emergency room with reddish wound on scalp a week ago. Patient planning for shunt removal, debridement and reinsertion of VP shunt. A and B Pre procedural CT Scan.

The fifth case was a 5-year-old boy had a wound on his stomach since 16 days before admission. Wound had bigger over time and tube appeared from the wound. The patient had a history of hydrocephalus and VP shunt installment when he was 4 months old. 4 years ago, the VP shunt was revised because of obstruction. The patient also had a history of consuming anti-tuberculous drugs 4 years ago. His body weight was 14 kg. The patient has received local infection control prior to his shunt insertion. Patient was planned & operated for shunt exteriorization. The patient had improved clinical outcome and was discharged from hospital care. The patient was followed up 6 months later with improved exposed site and a satisfactory result was reported from the parents (Fig. 5).

The sixth case was a 2-month-old baby with 3.2 kg of weight, who was admitted to the hospital complaining of exposed shunt. She suffered fever since the night before admission without vomiting and seizure. The patient was given symptomatic management following her admission to our hospital. She had a history of hydrocephalus surgery at 1.5 months old. The patient had improved clinical outcome and was discharged from hospital care. The patient was followed up 3 months later with improved exposed site and a satisfactory result was reported from the parents (Fig. 6).

3. Discussion

This study was an analysis of all cases treated in a period of 1 year from January to December 2018 with an inclusion criterion of history of exposed shunt of any age group. This study is a report of the clinical presentation and radiology examination before and after treatment. Clinical and radiology evaluation were performed in immediate post procedural period. A detailed clinical examination was performed to look for exposed shunt complication.

Radiological evaluation was done with shunt insertion or EVD. The patients came to the emergency room and neurosurgery clinic in our hospital with the same chief complaint; wound on the skin. Age distribution ranged from 2 months to 6 years old. Two patients had a history of seizure [6].

Shunt revision is more likely to be required if the catheter is placed extra ventricularly. Based on age, pediatric patients are more likely to require revision than adults. Shunt complications on pediatric has 4.22-fold greater odds of shunt revision compared to adults [10]. The six patients in this case series are pediatric patients. Factors that contribute to the VP shunt exposure are skin infection. Data analysis of 1015 patients in a single institution from 1970

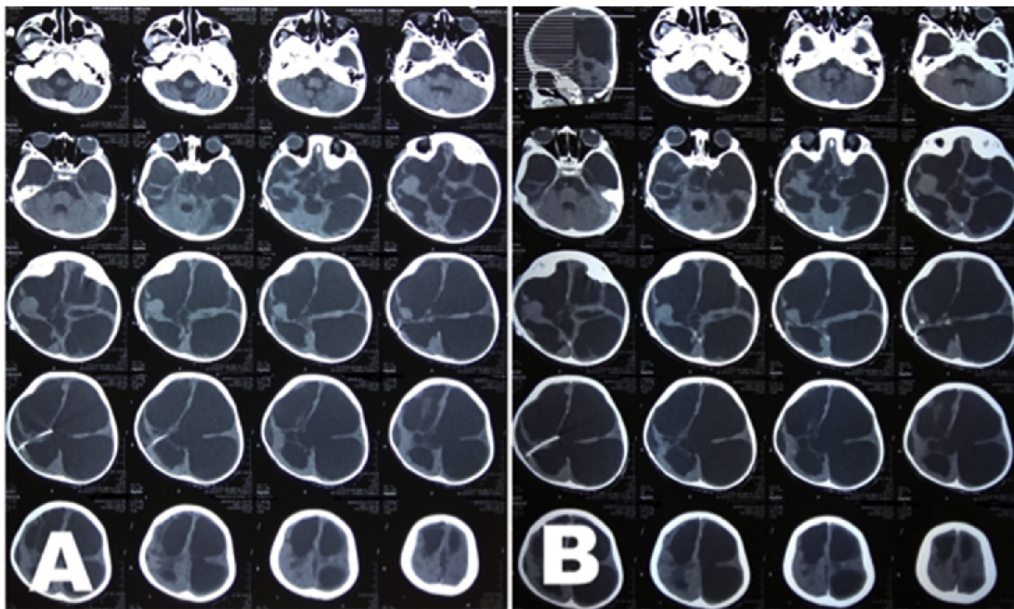


Fig. 5. The fifth case, a 5-year-old boy had a wound on his stomach since 16 days before admission. Wound had bigger over time and tube appeared from the wound. Patient was planned to shunt exteriorization. A and B Pre procedural CT Scan.

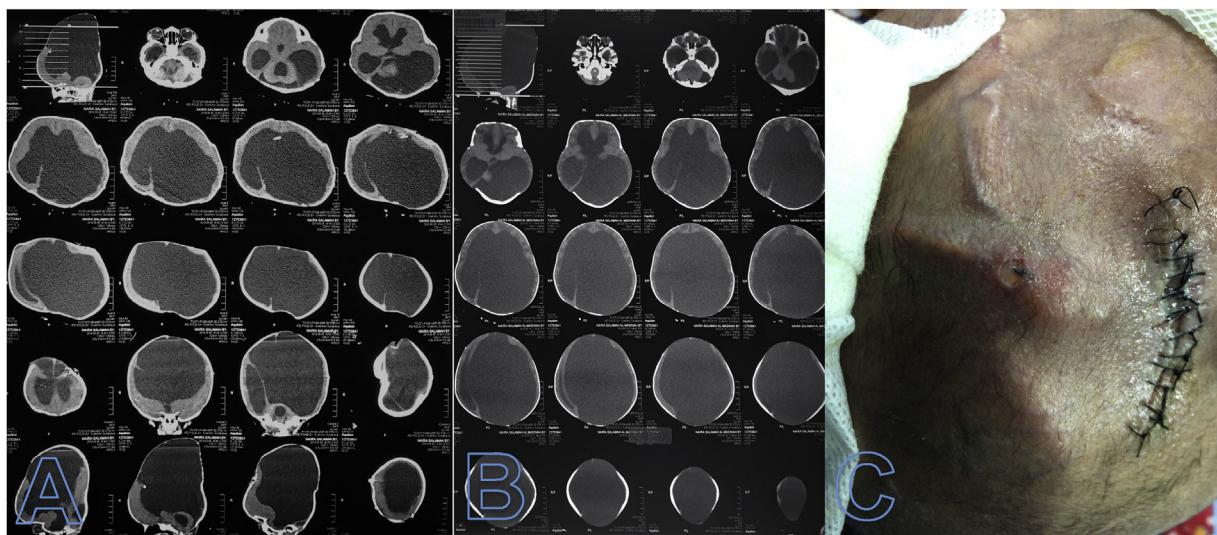


Fig. 6. The sixth case, a 2-month-old baby with 3,2 kg of weight was admitted to the hospital complaining with exposed shunt. A and B Pre procedural CT Scan; C. Pre procedural clinical image.

to 2010 indicated that age is a risk factor for shunt malfunction. Revision rates were significantly greater in pediatric (<17 years) patients (78.2% vs. 32.5%, $P < 0.001$), and shunt malfunction also tends to be high in patients with shunt revision. In these six patients which underwent shunt revision, long follow-up was considered an important thing to take note of [16]. Patient number four underwent shunt revision due to obstruction in early year of his life and four years later patient came with an exposed shunt. Younger age at first shunt placement (HR 1.6 [95% CI 1.1–2.1]) was associated with shunt malfunction [18].

The six patients in this case series were put on shunt under the age of 1 year. Some studies referred that the installation of shunts in early life increases the risk of shunt malfunction. Piatt and Carlson [19] in their study stated that patients who had shunt installed at age less than 1 month had higher risk for malfunction. Other study also supported this statement. Liptak and McDonald [20] stated that the risk for malfunction increased in installments at

less than 2 years. According to Park et al., infection rates were high at the beginning of the first 6 months of shunt installation while obstruction more commonly occurred later. In his study shunt malfunction was associated with the process of catheter degradation. This was hypothetically associated with fibrosis and tight adhesion with surrounding soft tissue. This adhesion inhibits sliding catheter to adjust to the growth of children although the mechanism is still unclear [21].

All six patients in this case series had poor nutritional status by their body weight. First patient with body weight of 15 kg (<50th percentile), second patient with body weight of 9 kg (<50th percentile), third patient with body weight of 5 kg (<50th percentile), fourth patient with body weight of 8 kg (<50th percentile), fifth patient with body weight of 15 kg (<3rd percentile), sixth patient with body weight of 7.5 kg (<50th percentile), and the sixth patient with a body weight of 3.2 kg (<50th percentile). Poor nutritional status might be an important risk factor for exposed shunt. We found

Table 1
CSF analysis.

Patient no.	Color	Clarity	Clot	Cell	PMN	MN	Nonne	Pandy	Glucose	Protein
1.	Colorless	Clear	Negative	0.005	75	25	Negative	Negative	62	7.44
2.	Yellowish	Clear	Negative	13	8.3	91.7	Positive	Positive	38	171
3.	Colorless	Clear	Negative	1	100	0			58	
4.										
5.	Colorless	Clear	Negative	0.02	42.9	57.1	Negative	Negative	80	15.59
6.										

that all the six patients had thin skin. Gyang M. et al. also presented a malnourished infant with post-infective hydrocephalus having a thin skin. It was presented that a subpericranial technique was used for a malnourished infant requiring ventriculoperitoneal shunt [22]. This technique involves making a scalp incision with pericranium taken in one layer with galea. If the galeal flap has been raised, a pericranial incision is made and a pericranial flap is raised. A subpericranial pouch is developed and a shunt pass through the it into the abdomen. The pericranial layer is closed. The galea and subcutaneous layer are also approximated, and a continuous subcuticular stitch is applied.

The first patient complained of fever for two months and the third patient also complained of the same fever. Direct exposure of shunt will increase the risk of infection. Fever may occur due to infection or from something else. Investigation should be done to confirm the cause of the fever. In all six patients there was no cerebrospinal fluid (CSF) leak or purulent discharge. A CT scan brain was performed to evaluate CSF and hydrocephalus conditions. Four of six patients underwent CSF analysis with results presented in table below (Table 1).

Four of six patient was analyzed for their CSF. CSF for patient number 2 had a raised cell numbers, PMN and MN and also positive result of Nonne and Pandy. The others showed CSF with normal limit. This infection will give an abscess or fistula. More fragile skin integrity makes the shunt easier to protrude out [23–25]. The shunt condition must be completed by the client. When protrusion occurs and the shunt is no longer functioning, a shunt revision must be done. CSF analysis and culture are needed to confirm infection [26]. If the infection is still obtained after antibiotic treatment, the shunt will be removed. CSF infection is a determining factor whether the shunt is maintained or released. If infection is persistent, shunt should be removed. If not, repair with a flap/flaps is required [26]. However, our study is still limited by the retrospective study design which was depended on the availability and accuracy of the data records, and we do not have a control arm so it may not be compared and be generalizable to a larger population of patients.

4. Conclusion

Shunt is still a routine therapy in the field of neurosurgery, although therapies such as endoscopic third ventriculostomy (ETV) have started to be done frequently. Complications such as exposed shunt are rare in the treatment of pediatric cases with hydrocephalus. We presented that exposed shunt is a rare complication (2.3% incidence rate), it might be caused by some of risk factor, such as the age when shunt was done and nutritional status. However, early diagnosis and treatment are solution to prevent further complications, especially infections. Subpericranial technique for shunt tunneling might be useful in preventing exposure of the shunt with associated morbidity factors.

Declaration of Competing Interest

The authors declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Funding

The authors received no financial support for the research, authorship and/or publication of this article.

Ethical approval

Ethical approval to report this case was obtained from The Hospital Research Ethics Committee of “Rumah Sakit Umum Daerah Dr. Soetomo” where the patient was admitted.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Wihasto Suryaningtyas, M.D: Conceptualization, Investigation, Resources, Writing – Original Draft, Supervision, Project Administration.

I G. M. Aswin R. Ranuh, M.D: Conceptualization, Writing – Original Draft, Writing – Review and Editing.

Muhammad Arifin Parenrengi, M.D., Ph.D.: Investigation, Resources, Writing – Original Draft.

Registration of research studies

Not applicable.

Guarantor

Wihasto Suryaningtyas, M.D.

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

- [1] H.L. Rekeate, Definition and classification of hydrocephalus, *Cerebrospinal Fluid Res.* 7 (Suppl. 1) (2010) S39, <http://dx.doi.org/10.1186/1743-8454-7-S1-S39>, Published 2010 Dec 15.
- [2] R.J. Lemire, Neural tube defects, *JAMA* 259 (1988) 558–562.
- [3] W. Norkett, D.G. McLone, R. Bowman, Current management strategies of hydrocephalus in the child with open spina bifida, *Top. Spinal Cord Inj. Rehabil.* 22 (4) (2016) 241–246.
- [4] M. Junaid, M. Ahmed, M.U. Rashid, An experience with ventriculoperitoneal shunting at keen's point for hydrocephalus, *Pak. J. Med. Sci.* 34 (3) (2018) 691–695.

- [5] A. Hamdan, Ventriculoperitoneal shunt complications: a local study at Qena University Hospital: a retrospective study, *Egypt. J. Neurosurg.* 33 (1) (2018).
- [6] L. Stratchko, I. Filatova, A. Agarwal, S. Kanekar, The ventricular system of the brain: anatomy and normal variations, *Semin. Ultrasound CT MRI* 37 (2) (2016) 72–83.
- [7] R.S. Tubbs, P. Oakes, I.S. Maran, C. Salib, M. Loukas, The foramen of Monro: a review of its anatomy, history, pathology, and surgery, *Childs Nerv. Syst.* 30 (10) (2014) 1645–1649.
- [8] M.M. Mortazavi, N. Adeeb, C.J. Griessenauer, H. Sheikh, S. Shahidi, R.I. Tubbs, R.S. Tubbs, The ventricular system of the brain: a comprehensive review of its history, anatomy, histology, embryology, and surgical considerations, *Childs Nerv. Syst.* 30 (1) (2013) 19–35.
- [9] S. Krishnamurthy, J. Li, New concepts in the pathogenesis of hydrocephalus, *Transl. Pediatr.* 3 (3) (2014) 185–194.
- [10] M. Paff, D. Alexandru-Abrams, M. Muhonen, W. Loudon, Ventriculoperitoneal shunt complications: a review, *Interdiscip. Neurosurg.* 13 (2018) 66–70.
- [11] H. Tully, W. Dobyns, Infantile hydrocephalus: a review of epidemiology, classification and causes, *Eur. J. Med. Genet.* 57 (8) (2014) 359–368.
- [12] M. Kiefer, A. Unterberg, The differential diagnosis and treatment of normal-pressure hydrocephalus, *Arztebl. Int.* 109 (1–2) (2012) 15–25, quiz 26.
- [13] K. Kahle, A. Kulkarni, D. Limbrick, B. Warf, Hydrocephalus in children, *Lancet* 387 (10020) (2016) 788–799.
- [14] M.R. Del Bigio, D.L. Di Curzio, Nonsurgical therapy for hydrocephalus: a comprehensive and critical review, *Fluids Barriers CNS* 13 (1) (2015).
- [15] P. Pan, Outcome analysis of ventriculoperitoneal shunt surgery in pediatric hydrocephalus, *J. Pediatr. Neurosci.* 13 (2) (2018) 176–181.
- [16] G.K. Reddy, P. Bollam, G. Caldito, Long term outcomes of ventriculoperitoneal shunt surgery in patients with hydrocephalus, *World Neurosurg.* (2014) 404–410, 01.96.
- [17] R.A. Agha, C. Sohrabi, G. Mathew, T. Franchi, A. Kerwan, N. O'Neill, for the PROCESS Group, The PROCESS 2020 guideline: updating consensus preferred reporting of CasE series in surgery (PROCESS) guidelines, *Int. J. Surg.* 84 (2020) 231–235.
- [18] J. Riva-Cambrin, J. Kestle, R. Holubkov, J. Butler, A. Kulkarni, J. Drake, et al., Risk factors for shunt malfunction in pediatric hydrocephalus: a multicenter prospective cohort study, *J. Neurosurg. Pediatr.* (2016) 382–390.
- [19] J.H. Piatt Jr., C.V. Carlson, A search for determinants of cerebrospinal fluid shunt survival: retrospective analysis of a 14-year institutional experience, *Pediatr. Neurosurg.* 19 (1993) 233–241.
- [20] G.S. Liptak, J.V. McDonald, Ventriculoperitoneal shunts in children: factors affecting shunt survival, *Pediatr. Neurosci.* 1986 (12) (1985) 289–293.
- [21] M.K. Park, M. Kim, K.S. Park, S.H. Park, J.H. Hwang, S.K. Hwang, A retrospective analysis of ventriculoperitoneal shunt revision cases of a single institute, *J. Korean Neurosurg. Soc.* 57 (5) (2015) 359–363.
- [22] G.M. Bot, N.J. Ismail, B. Usman, D.J. Shilong, J.O. Obande, S.O. Aliu, B.B. Shehu, Subpericranial shunt valve placement: a technique in patients with friable skin, *Childs Nerv. Syst.* 30 (8) (2014) 1431–1433.
- [23] S. Kobayashi, T. Ishikawa, T. Mutoh, K. Hikichi, A. Suzuki, A novel technique for ventriculoperitoneal shunting by flat panel detector CT-guided real-time fluoroscopy, *Surg. Neurol. Int.* 3 (1) (2012) 119.
- [24] K.J. Jeremiah, C.L. Cherry, K.R. Wan, J.A. Toy, R. Wolfe, R.A. Danks, Choice of valve type and poor ventricular catheter placement: modifiable factors associated with ventriculoperitoneal shunt failure, *J. Clin. Neurosci.* 27 (2016) 95–98.
- [25] M. Paff, D. Alexandru-Abrams, M. Muhonen, W. Loudon, Ventriculoperitoneal shunt complications: a review, *Interdiscip. Neurosurg.* 13 (2018) 66–70.
- [26] O. Akdag, Management of exposed ventriculoperitoneal shunt on the scalp in pediatric patients, *Childs Nerv. Syst.* 34 (6) (2018) 1229–1233.

Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.