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Case Series and literature review: Intraventricular Cysts and Hydrocephalus Undergoing Cystoventriculostomy and VP Shunt Procedures

Farhad Bal'afif[1*], Donny Wisnu Wardhana[1], Tommy Nazwar Alfandy[1], Muhammad Ridwan[2], Mustofa Mustofa[2]

Division of Neurosurgery Department of Surgery Brawijaya University, Saiful Anwar Hospital Malang Indonesia[1]
Department of Surgery Brawijaya University Malang Indonesia[2]

Corresponding Author: 1*

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ABSTRACT

Intraventricular cysts, also known as intraventricular arachnoid cysts or intraventricular cerebrospinal fluid cysts, are rare cases and usually show asymptomatic to symptomatic intracranial Pressure elevation. Intracranial intraventricular cyst is a collection of clear fluid and does not resemble cerebrospinal fluid (CSF) that occurs in the ventricles. Intraventricular cysts are generally treated with open surgery such as peritoneal shunting, microfenestration with partial excision, endoscopic fenestration, stereotactic aspiration cystocisternostomy, and cystoventriculostomy. Endoscopic cystoventriculostomy has become common in recent years, due to its successful operation without touching the basal cistern in most cases with minimal complications. Objectives of this study reports a case series and literature review: patients with intraventricular cysts and hydrocephalus who underwent cystoventriculostomy and VP shunt procedures. We prospectively identified patients with intraventricular cysts who developed hydrocephalus who required a cystoventriculostomy procedure. Patients were followed up for development of hydrocephalus, need for a VP shunt, and length of stay in the intensive care unit. The first case was a 1 year old child with a history of communicating hydrocephalus with a VP shunt, with complaints of vomiting, restlessness, and difficulty sleeping. The results of the head MRI with contrast showed that there were multiloculated cystic lesions in the III and IV ventricles. The second case was a 79-year-old man with complaints of seizures and a history of decreased consciousness. A CT scan of the head revealed a cystic lesion with rim of calcification in the left frontal lobe. Both patients underwent endoscopic fenestration with good outcome in both patients. Although the arachnoid cyst is a relatively benign pathological entity, preemptive therapy can be managed with endoscopically guided fenestration of the cyst wall. Endoscopic fenestration is the technique of choice because the risks and complications associated with open craniotomy and fenestration are less.

1. INTRODUCTION

Intraventricular cysts are classified into two groups, the first group includes cysts originating in the central nervous system such as arachnoid cysts, ependymal cysts, loculated hydrocephalus, choroid plexus cysts, and porencephalic cysts. The second group includes cysts resulting from the intrusion of non-neural tissue into the neuro-axis, usually in the midline, such as colloid cysts and Rathke's cleft cysts. Intraventricular cyst, is a rare case and usually shows no symptoms to show symptoms of increased intracranial [1].

Intracranial intraventricular cysts are clear and colorless collections of fluid resembling Cerebrospinal Fluid (CSF) which account for approximately 0.5-1% of all intracranial lesions. The most common intraventricular cysts are arachnoid cysts, where arachnoid cysts occur within the arachnoid membrane and in the subarachnoid space of the main cerebral fissure and arachnoid cisterna, most commonly within the Sylvian fissure, while other locations include the quadrigeminal tectum, sellar and suprasellar regions, cerebellopontine angle, and posterior infratentorial midline cisterna. All three ventricular arachnoid cysts are even rarer and to our knowledge, only 7 cases have been reported to date [1], [2].

Arachnoid cysts can occur as a result of developmental defects, inflammatory processes, trauma, bleeding, chemical irritation, or tumors caused by CSF sequestration. While cysts are usually found as incidental asymptomatic findings, some of them can develop, can compress surrounding structures, and therefore become symptomatic. Intracranial arachnoid cyst is a relatively common benign congenital lesion occurring in about 1.1% of the population worldwide. This disorder is more common in children than in adults. Most cases are sporadic and asymptomatic. A small number of patients present with neurologic symptoms [3], [4].

Arachnoid cysts are generally treated with open surgery such as peritoneal shunting, microfenestration with partial excision, endoscopic fenestration, stereotactic aspiration cystocisternostomy, and cystoventriculostomy. Endoscopic cystoventriculostomy has become common in recent years, due to its successful operation without touching the basal cistern in most cases with minimal complications. The complications mentioned in the literature are late onset chronic SDH and very late onset brain abscess [5]. However, there is still some controversy regarding the best surgical treatment for arachnoid cysts [6], [7].

We present a case series of intracranial arachnoid cysts and hydrocephalus, which were treated with cystoventriculostomy and VP shunt.

2. Methods

Setting:

This research was conducted at the Department of Neurosurgery, RSUD Dr. Saiful Anwar Malang in August 2021. Saiful Anwar Hospital cooperates with Universitas Brawijaya, Faculty of Medicine. The division in this study is Neurosurgery.

Research Population

Neurosurgery patients undergoing treatment in the intensive care unit of RSUD Dr. Saiful Anwar with intraventricular cysts and hydrocephalus in the period August 2021 requiring a cystoventriculostomy procedure.

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Research Sample
The selection of the research sample was carried out by purposive sampling technique.

Design:
We conducted a retrospective review of two cases of patients with intraventricular cysts and hydrocephalus who were admitted to the intensive care unit of RSUD Dr. Saiful Anwar Malang in the period of August 2021 who requires a cystoventriculostomy procedure. Patients were followed up for development of hydrocephalus, need for a VP shunt, and length of stay in the intensive care unit.

Data collection
The collection of anamnestic, clinical, therapeutic and follow-up data for each patient was obtained from the electronic medical record (RME). Clinical data in the form of age, medical history, and risk factors were collected.

Ethical Approval
We got consent from the head of RSUD Dr. Saiful Anwar Malang to publish data and maintain patient anonymity.

3. Case presentation

Case 1
A 1 year old child with a history of communicating hydrocephalus with a VP shunt (Figure 1). The patient came with complaints of vomiting, seemed restless, and had trouble sleeping. The patient also had a history of congenital heart disease, namely moderate L-R secundum ASD, moderate PMO VSD, mild pulmonary regurgitation, and moderate pulmonary hypertension. Physical examination revealed a sunset eye phenomenon (+/+), dry and clean VP shunt surgery scars, and head circumference of 48 cm (figure 1). The results of the head MRI with contrast are multiloculated cystic lesions in the III and IV intraventricular, suspected intraventricular arachnoid cyst causing severe obstructive hydrocephalus at the level of the third ventricle on VP shunt and agenesis of the septum pellucidum, dysgenesis of the corpus callosum. The patient was programmed for endoscopic cyst fenestrations surgery, ETV, VP shunt kocher dextra low press, and aff VP shunt keen dextra (figure 2).

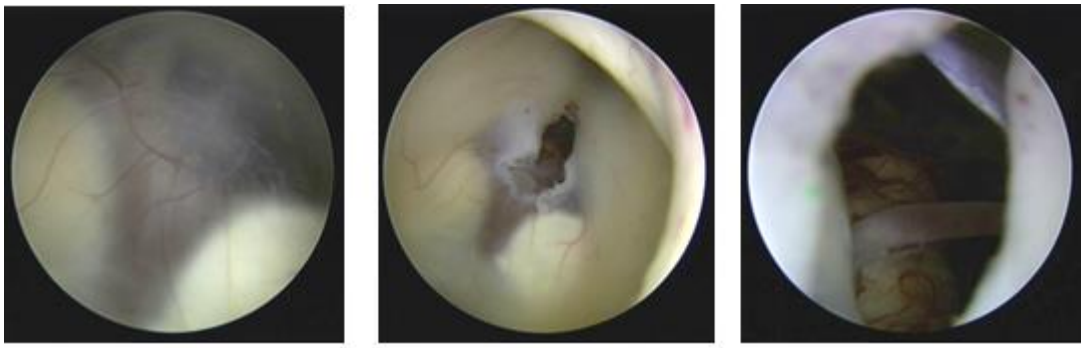

Figure 1. MRI of the head of case 1. There was a multiloculated cystic lesion in the III and IV intraventricular, with the appearance of an arachnoid cyst. There is also agenesis of the septum pellucidum and dysgenesis of the corpus callosum.

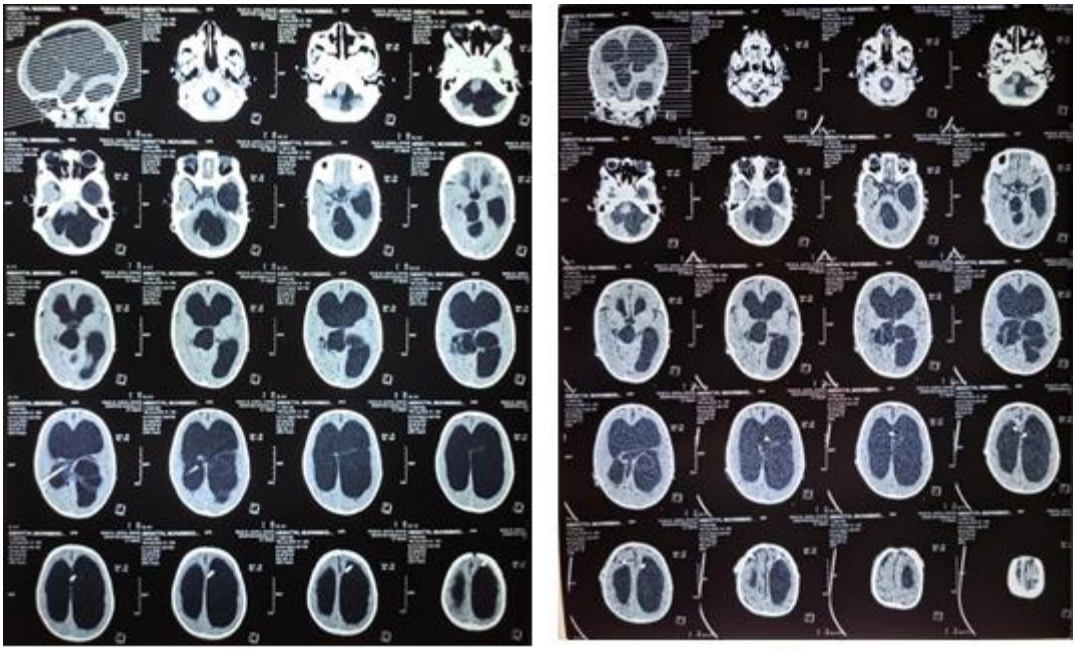

Figure 2. Case 1 of CT scan representation. (A) preoperative CT scan, (B) postoperative CT scan

Case 2

The patient is a 79 year old male with complaints of seizures. Seizure 1 time for 1 minute. The patient is unconscious after the seizure. Previously, the patient had a history of loss of consciousness. Previous medical history, namely pulmonary Ca on treatment and coronary heart disease with a history of CABG. A CT scan of the head revealed a cystic lesion with rim of calcification in the left frontal lobe, chronic lacunar infarction of the right internal capsule genu, left thalamus, senile brain atrophy and moderate hydrocephalus (figure 3 & 4). The patient was programmed for endoscopic cyst fenestration + low pressure VP shunt.

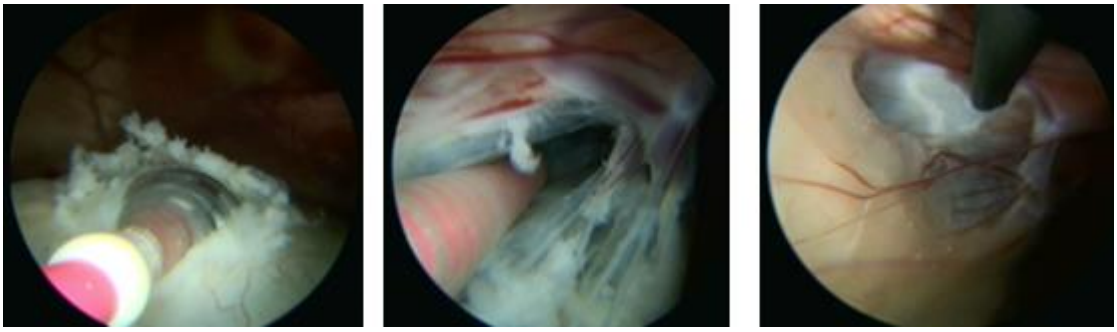

Figure 3. Result of head CT scan of case 2. There is a cystic lesion with rim of calcification in the left frontal lobe. Lacunar infarcts were also seen in the right internal capsule genu and left thalamus.

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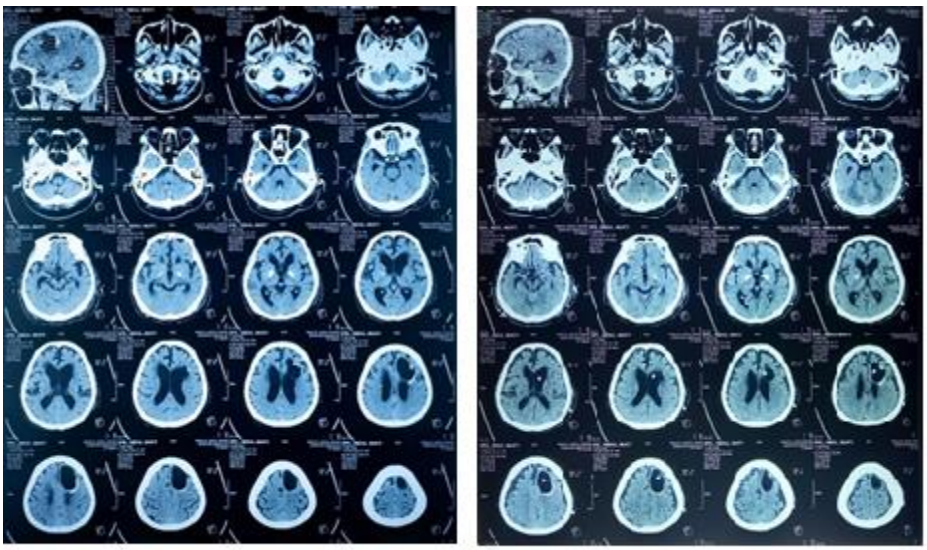

Figure 4. Case 2 of CT scan representation. (A) preoperative CT scan, (B) postoperative CT scan.

Case	History	Age	Procedures	Findings
1	hydrocephalus with a VP shunt	1 year old	endoscopic cyst fenestrations surgery	There was a multiloculated cystic lesion in the III and IV intraventricular, with the appearance of an arachnoid cyst. There is also agenesis of the septum pellucidum and dysgenesis of the corpus callosum
2	moderate hydrocephalus	79 year old	endoscopic cyst fenestration + low pressure VP shunt	Paraventricular arachnoid cystic lesion with rim of calcification in the left frontal lobe. Lacunar infarcts were also seen in the right internal capsule genu and left thalamus

4. Discussion

An arachnoid cyst is a sac filled with cerebrospinal fluid that lies between the brain or spinal cord and the arachnoid membrane, one of three membranes that cover the brain and spinal cord. Arachnoid cysts occur in about 1.4% in adults, whereas in children the prevalence is between 1% and 3%. The male to female ratio is about 3:1. These cysts are divided into primary and secondary cysts. Symptoms: headache (66%), dizziness,

nausea, vomiting, bad mood, altered mental status, ataxia, seizures, and hearing loss [1], [2], [8], [9].

In general, the location is 70% supratentorial (sylvian > intraventricular, suprasellar), 25% spinal and 5% infratentorial. Other locations include the quadrigeminal plate, sellar area, and convexity. The majority of middle cranial fossa cysts (about 50%) are reported to be of moderate size. The location of the middle cranial fossa may be explained by meningeal maldevelopment, the arachnoid covering of the temporal and frontal lobes failing to fuse when the Sylvian fissure is formed early in fetal life, thus creating a non-communicating fluid compartment completely surrounded by the arachnoid membrane. They occupy the anterior and middle portions of the temporal fossa and often displace the ends of the temporal lobes posteriorly, superiorly, and medially [3].

Arachnoid cysts can be congenital (primary) or acquired (secondary). When there is no evidence of a traumatic or inflammatory origin, the cyst is considered congenital. The majority of arachnoid cysts are congenital. Initially, two theories were proposed regarding the etiology of arachnoid cysts. The first theory is the "intra-arachnoid cyst theory" proposed by [10], in which the cyst is believed to be the result of division and duplication of the arachnoid membrane. The second is Robinson's “subarachnoid cyst theory” [11], which states that middle cranial fossa cysts form in response to temporal lobe agenesis. [8], [10], [11]. Currently, several theories have been proposed to explain the etiology of arachnoid cysts such as agenesis of the corpus callosum, developmental abnormalities of the arachnoid, malformations of the cerebral venous system, or CSF flow abnormalities [3], [12].

Primary (congenital) arachnoid cyst is a developmental disorder in which division or duplication of the primitive arachnoid membrane early in embryonic life causes a collection of fluid such as cerebrospinal fluid. In acquired cysts the intracystic fluid may be stained with hemosiderin or may contain inflammatory cells with a possible gradient. In congenital arachnoid cyst walls contain arachnoid cells connected to unchanged arachnoid material and in secondary cysts, the CSF loculation is surrounded by arachnoid scar tissue. Secondary arachnoid cysts may result from CSF sequestration due to inflammation or following traumatic processes, bleeding, chemical irritation, and tumors. In secondary cysts, most of which occur after inflammation or brain trauma, the CSF loculation is surrounded by arachnoid scar tissue. The etiology of primary arachnoid cyst formation is uncertain, but is in theory predominately due to abnormal cleavage of the arachnoid during embryogenesis. Several syndromes have been associated with arachnoid cysts: Aicardi syndrome; mucopolysaccharidosis; Acrocallosal syndrome, Marfan syndrome and Chudley-Mullough syndrome. The etiology of secondary arachnoid cysts is head injury, meningitis,/infection, tumor, or complications from brain surgery [3].

The clinical manifestations of arachnoid cysts are variable and often non-specific. In a patient with an arachnoid cyst, neurologic signs and symptoms reflect their size, anatomy, distribution and impact on CSF flow [3]. In this case series, the first case of a 1-year-old pediatric patient presented with vomiting, restlessness, and difficulty sleeping. On physical examination, we found a sunset eye phenomenon (+/+), dry and clean VP shunt surgery scars, and head circumference of 48 cm. In the second case, a 79-year-old male patient with complaints of seizures. Seizure 1 time for 1 minute. The patient is unconscious after the seizure. Previously, the patient had a history of loss of consciousness. Intraventricular arachnoid cysts are usually asymptomatic. When arachnoid cysts are symptomatic, patients may present with nausea, vomiting, headache, focal neurologic deficits, seizures and symptoms of obstructive hydrocephalus. Similar symptoms were found in our case such as headache and convulsions [1], [2].

Although most of the lesions are asymptomatic and remain stable on serial imaging studies, arachnoid cysts

can enlarge and become symptomatic secondary to the crowding effect. The mechanism of enlargement of the arachnoid cyst may be related to: (1) secretion of fluid from the cyst wall, (2) an increase in the intracystic osmotic gradient that promotes fluid infiltration through the cyst wall or (3) a spherical mechanism with valves in which cerebrospinal fluid is trapped within the cyst during periods of elevation. transient intracranial pressure, such as during the Valsalva maneuver. Middle fossa arachnoid cysts can present with many signs and symptoms, including headache, increased intracranial pressure, macrocephaly, focal neurological deficits, seizures, cognitive decline, endocrine dysfunction and acute or chronic subdural hematoma after minor trauma [1], [3].

When small arachnoid cysts are usually asymptomatic, but large cysts in the supratentorial, suprasellar, and posterior fossa can cause hydrocephalus. Large middle cranial fossa cysts may be associated with seizures, headache, or, rarely, hemiparesis. The most common signs and symptoms are increased intracranial pressure, craniomegaly, and developmental delay. They are usually seen in cases of large supratentorial cysts, but can also be caused by suprasellar or posterior fossa cysts associated with obstructive hydrocephalus. Hydrocephalus is estimated to be present in 30-60% of patients with arachnoid cysts [3].

Differential diagnosis among primary intracranial cysts is essential for adequate treatment. Some symptomatic cysts, such as neurenteric and epidermoid/dermoid, must be surgically removed, including the cystic wall and contents to avoid recurrence [13]. The differential diagnosis of arachnoid cysts includes intraventricular (eg colloid cysts), intraparenchymal (eg parasitic infections, cystic metastases), porencephalic cysts, craniopharyngiomas, holoprosencephalies, certain forms of agenesis of the corpus callosum, defects in the hemispheral cleavage, and the Dandy–Walker complex (posterior fossa cyst) [3], [8].

Diagnostic evaluation should include early identification of intracranial arachnoid cysts, detection of mass effect, determination of the type of communication between the cyst and the subarachnoid space and recognition of the presence, location and severity of obstructive hydrocephalus and cisternal block. Diagnostic tools for arachnoid cysts are computerized tomogram (CT) with contrast studies, magnetic resonance imaging (MRI), radioisotope scintigram and cine phase contrast MR imaging. On CT, the arachnoid cyst was observed as an extraaxial cyst with CSF density, without any contrast enhancement. Contiguous calvarial remodeling is common, due to hypoplasia of the adjacent brain parenchyma, especially in the middle cranial fossa arachnoid cyst. 3 MRI allows assessment of the cyst margin, its contents, and the extent of the lesion. The MRI signal is similar to that of CSF on T1- and T2-weighted imaging with no enhancement of gadolinium with normal signals. Brookes et al. have demonstrated pulsatile movement of cerebrospinal fluid within cysts on MRI [3], [13].

Several treatment options are currently optional for the management of arachnoid cysts, such as craniotomy with resection of the cyst wall and contents, stereotactic aspiration or fenestration of the cyst cavity, and neuroendoscopic fenestration or shunting procedures. The most commonly performed are craniotomy and shunting, craniotomy using fenestrated endoscopy. The neurosurgeon makes a small cut (incision) near the site of the arachnoid cyst. Then the neurosurgeon places the endoscope and looks for the cyst. Then make a hole in the cyst, After the arachnoid cyst has been opened, the fluid in it flows into other areas of the brain that contain cerebrospinal fluid so that it will be reabsorbed. In addition, treatment can be done with shunting. A shunt is a small tube (catheter) that drains cerebrospinal fluid from one place in the body to another [14-16].

If the cyst is located in an indeterminate area and resection of the cyst from brain tissue is possible without damage, total excision of the cyst, including its wall and contents, is the most appropriate treatment for

symptomatic lesions. Each of these techniques, however, has been associated with postoperative complications. Shunting of arachnoid cysts, although simple, is associated with shunt infection, failure and overdrainage with revision rates of 30% or more. To avoid shunt dependence and associated complications, craniotomy and cyst fenestration have been tried but with higher morbidity (10-15%) and mortality (1%). With the introduction of endoscopy, the risks and complications associated with open craniotomy and fenestrations have been minimized [14- 16].

Endoscopic cyst fenestration has gained popularity with reported excellent results. Magnetic resonance imaging – or computed tomography-guided endoscopy can be used, access to the cyst is obtained through coagulation of the outer cyst membrane, and cystocisternostomies are performed. Access neuroendoscopy has also been used as an adjunct to open microsurgical fenestrations. As this technique gains exposure, minimally invasive fenestration may become the preferred surgical technique for accessible cysts [17], [18].

The goals of surgical management for arachnoid cysts are to communicate the interior of the cyst with the anatomic corridor of CSF flow or the application of a shunt from the cyst or ventricular system to another cavity where CSF can be absorbed. Surgical techniques for the management of arachnoid cysts include microsurgery, excision, cystperitoneal shunting, endoscopic ventriculocystostomy or ventriculocystocistenostomy, stereotactic cyst-ventricular shunting, and stereotactic intracavitary irradiation. The choice of the most appropriate surgical approach is still debated and many surgical procedures for the treatment of arachnoid cysts have been recommended and it is still controversial as to which method is best. Harsh et al stated that all mass-effect arachnoid cysts, especially in children, should be treated. The potential for inhibiting adjacent brain development and function, and for cyst rupture, intracystic haemorrhage, or subdural haemorrhage causing sudden severe neurologic damage outweighs the risk of operative treatment [3], [13], [19].

Arachnoid cysts have a 33% recurrence rate, suggesting that cyst-peritoneal shunting is necessary in intraventricular arachnoid cysts to prevent recurrence. Endoscopy is better for the management of suprasellar arachnoid cysts than microsurgical and shunting procedures. Currently, endoscopic cyst fenestration provides the advantage of controlling triventricular dilatation and avoiding shunt dependence in the follow-up period [19].

Fenestration of the cyst or resection via craniotomy carries a potential risk of complications such as neurologic deficits, meningitis, subdural collection, and epileptic seizures. As a result, the endoscopic approach has recently become popular in many areas for the treatment of arachnoid cysts, as it is less invasive and helps avoid complications associated with shunting [2]. Neuroendoscopy, however, is not a risk-free procedure. The risks of neuroendoscopy include bleeding, with difficulty controlling bleeding, infection, and increased intracranial pressure from too much irrigation without proper ventilation. Endoscopic treatment appears to be new, safe and an alternative to open surgery and shunting in the management of arachnoid cysts. The results in the literature are comparable to traditional methods with the advantage of a low incidence of complications [20].

Hoffman [21], recommend the transcallosal approach based on the fact that it allows communication between the cyst and the ventricles to be established. While a transcallosal approach via craniotomy can achieve fenestration and/or resection of the cyst, the open approach can also result in damage to important structures including vascular injury, disconnection syndrome, fornix injury and subcortical core damage. On the other hand, [22] reported that 67% of patients with intracranial arachnoid cysts operated on by craniotomy required subsequent cyst shunting [2].

Colloidal cysts are benign growths that are usually located in the third ventricle and at or near the foramen Monroe, which is on the anterior aspect of the third ventricle of the brain. Colloidal cysts can cause a variety of symptoms, including headaches, diplopia, memory problems, and vertigo. Rarely colloid cysts have been cited as a cause of sudden death. When colloid cysts are symptomatic, they most often cause headache, nausea, and vomiting secondary to obstructive hydrocephalus. Obstructive hydrocephalus is precipitated by blocking the exit of cerebrospinal fluid (CSF) from the lateral ventricles at the foramen Monro, which connects the lateral and third ventricles. Most of these patients present with symptoms of hydrocephalus, including headache, nausea, vomiting, blurred vision, gait ataxia, and cognitive decline. Notably, a minority of patients present with symptoms of acute obstructive hydrocephalus and progress to death despite rapid CSF diversion and appropriate supportive care. Estimates of the risk of rapid clinical deterioration due to acute hydrocephalus in patients with colloid cysts vary widely, from 3% to 35%, with an associated 5%-38% risk of death. Historically, these patients were often described as having "sudden deterioration and death," but several recent studies using modern neuroimaging techniques have shown that nearly all patients with acute obstructive hydrocephalus have progressive symptoms before a sharp clinical deterioration [23], [24].

The term “multiloculated hydrocephalus” denotes the presence of an isolated CSF compartment within the ventricular system that may enlarge despite a functioning shunt system. The presence of an intraventricular septum or obstruction between the site of CSF production and the ventricular end of the catheter can prevent CSF outflow, leading to accumulation of fluid into the compartment. Other terms used to denote this condition are multicompartmental, multicystic, loculated, or complex hydrocephalus [25].

Based on the location of the obstruction and subsequent fluid accumulation, the anatomy and radiographic features of multiloculated hydrocephalus are variable and can be classified as follows: (1) Multiple intraventricular septations; (2) Isolated lateral ventricle/Unilateral hydrocephalus; (3) Entrapped temporal horn; (4) Isolated fourth ventricle; and (5) Expanding cavum septi pellucidi (CSP)/cavum Vergae (CV) [25].

The etiology and pathogenesis of multiloculated hydrocephalus is traditionally due to insults occurring during the neonatal period such as intraventricular hemorrhage and neonatal meningitis. However, other conditions may play an important role as an etiologic factor, such as shunt-associated infection, overdrainage, direct ependymal trauma during catheter insertion, head injury and intracranial surgery [25].

Ventriculitis is inflammation of the ependymal lining of the cerebral ventricles, usually secondary to infection. It has other names, such as ependymitis, ventricular empyema, pyocephalus, and pyogenic ventriculitis. Early diagnosis is very important for proper treatment. This is of particular concern in patients with external ventricular drainage (EVDs) or intraventricular shunts [26], [27].

Chronically, ventricular septations may develop, resulting in multiloculated hydrocephalus, which worsens the prognosis and is more common in bacterial infections. If left untreated, ventriculitis can lead to poor neurology, hydrocephalus, and death. Early recognition and treatment is very important. Because of the risk of recurrence and hydrocephalus, long-term follow-up is recommended. The ventricles and choroid plexus can serve as reservoirs of infection, even when a lumbar puncture produces sterile cultures. The definitive treatment is surgery, but its treatment remains controversial [26], [27].

In this case, a VP shunt was also performed in both the first and second cases. The VP shunt is effective in the treatment of hydrocephalus associated with arachnoid cysts. However, these shunts often require further revision. It should be noted, however, that shunting of cysts, which is technically difficult, differs from shunting for cyst-associated hydrocephalus. Despite all these known risks, a VP shunt can be useful and

sometimes unavoidable when hydrocephalus and its clinical findings persist after cyst resection or fenestration [2].

5. Conclusion

Although the arachnoid cyst is a relatively benign pathological entity, therapy in children, or in patients with symptomatic lesions regardless of age, can be managed by endoscopically guided cystoventriculostomy into the ventricular system or cyst spaces containing fluid resembling CSF. Proper patient selection, preoperative endoscopic trajectory planning, use of frameless navigation, and advances in endoscopic lens technology and light intensity combine to make this a relatively safe procedure with excellent outcomes.

Limitations: this study was limited by its retrospective and the small sample size nature, and unavailability of follow-up data. Future research with prospective enrollment and longer follow-up is needed.

What is known about this topic

· Arachnoid cysts or intraventricular cerebrospinal fluid cysts, are rare cases and usually show asymptomatic to symptomatic intracranial elevation, and the etiology remains unknown;

· Despite this, ventricular arachnoid cysts are rare, because there is no arachnoid tissue in the ventricles.

· The treatment is not specific and treated with open surgery.

What this study adds

· Arachnoid cyst events presents VP shunt procedures.

· Endoscopically guided cystoventriculostomy into the ventricular system can be managed regardless of age

Competing interests

The authors declare no competing interests.

Authors' contributions

Farhad Bal'afif: conception, literature review, analysis, data collection, writing-review and editing; Tommy N Alfandy: conception, software, writing review and editing; Donny W Wardhana: conception, software, writing-review and editing; Muhammad Rdiwan: conception, software, writing-review and editing; Mustofa Mustofa: conception, methodology and supervision; All the authors have read and agreed to the final manuscript

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