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Case Report

Intraparenchymal pericatheter cyst as an indicator of ventriculoperitoneal shunt malfunction: A case-based update

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ABSTRACT

Background: Intraparenchymal pericatheter cysts (IPCs) are a rare ventriculoperitoneal shunt (VPS) complication, with only a few cases recorded in the literature.

Case Description: We report a 22-year-old woman admitted with headache, papilledema, vision loss, and a history of leukemia. Lumbar puncture revealed idiopathic intracranial hypertension (IIH). Three months after VPS implantation, she was readmitted with headache and worsening of visual impairment. CT evidenced a IPC with perilesional edema. Intraoperatively, a shunt revision and cyst drainage were opted for. We present a discussion and literature review on this unique complication of VPS, with emphasis on management.

Conclusion: It is important to understand and consider IPCs as complications of VPS surgery, including in adult patients and IIH cases.

Keywords: Catheter obstruction, Cerebrospinal fluid edema, Idiopathic intracranial hypertension, Intraparenchymal pericatheter cyst, Ventriculoperitoneal shunt

INTRODUCTION

Ventriculoperitoneal shunt (VPS) is the main surgical intervention for communicating or obstructive hydrocephalus and idiopathic normal-pressure hydrocephalus. It can also be performed in idiopathic intracranial hypertension (IIH) when there is no response to medical therapy or there is visual dysfunction worsening. Although standard in several conditions, the procedure shows a considerable rate of complications (~23.8% in adults).[7]

Intraparenchymal pericatheter cyst (IPC) is a rare complication of VPS, occurring mainly in children. To date, only 35 cases have been reported in the literature, of which, 14 were in adults, and four were in IIH patients. The aim of this article is to present a case of IPC formation in an adult, as well as to review the literature on the subject.

CASE PRESENTATION

A 22-year-old woman with a history of leukemia presented with progressive headache, papilledema, and visual loss at admission. Magnetic resonance imaging (MRI) was normal [Figure 1]; however, lumbar puncture with cerebrospinal fluid (CSF) pressure measurement confirmed the diagnosis of IIH (pseudotumor cerebri), requiring treatment with VPS [Figures 1 and 2].

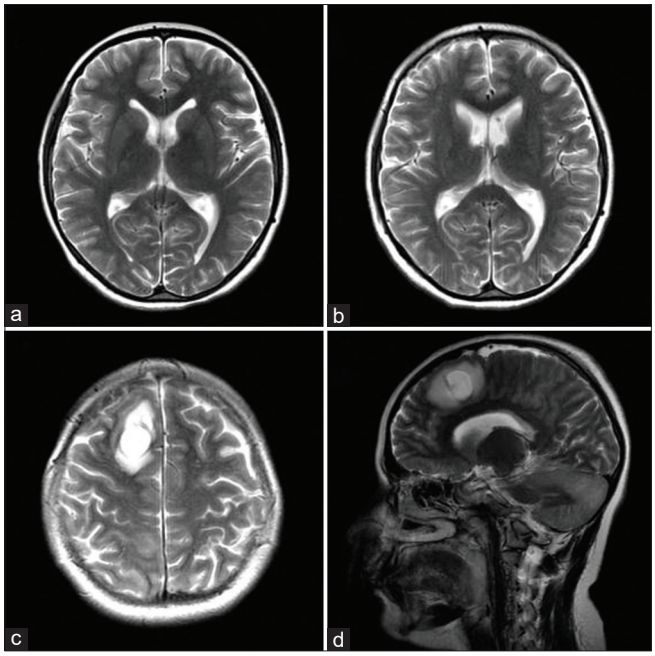

Figure 1: Imaging comparison. MRI at admission (before VPS surgery): (a) axial T2 view demonstrating no hydrocephalus. The lumbar puncture confirmed the diagnosis of idiopathic intracranial hypertension. MRI at revaluation (3 months later): (b) lower axial T2, (c) upper axial T2, and (d) parasagittal T2W1 views demonstrating the catheter pathway with an intraparenchymal pericatheter cyst and perilesional edema.

The patient was readmitted 3 months later due to the recurrence of headache symptoms and visual impairment. Computed tomography (CT) [Figure 2] showed a parenchymal lesion around the ventricular bypass catheter with surrounding edema, but without hydrocephalus (never present in this case). MRI [Figure 1] revealed a round-shaped lesion, with content isointense to CSF, and a thin nonenhancing wall. These findings were compatible with the diagnosis of IPC. Abdominal CT was benign, without evidence of distal causes of obstruction.

VPS revision was performed, and a proximal obstruction identified [Figure 3], then managed by catheter replacement and subsequent cyst drainage. Postoperative CT [Figure 2] revealed cyst regression and an intracystic hemorrhage. At 3-month follow-up, significant improvement in headache and visual impairment was observed, with a normal neurological examination and no surgical complications.

DISCUSSION AND LITERATURE REVIEW

VPS is a very common procedure, used mainly for treating hydrocephalus or idiopathic normal-pressure hydrocephalus. However, it is also an alternative for IIH, particularly in refractory cases or those involving visual deterioration.[3] In the literature, hydrocephalus was the main cause for VPS surgery (31 of 35 published cases, ~91%). Despite being frequent, VPS has a significant rate of complications, commonly divided into foreign body related (e.g., migration, infection, fibrosis, and allergic reaction), abdominal (e.g., pseudocyst) and surgical complications (e.g., overdrainage), and those resulting in hydrocephalus (e.g., obstruction and disconnection).[21]

IPCs, however, are very rare complications of VPS. Although they can also appear in Ommaya reservoirs, only VPS cases were considered in this review. Furthermore, only reports with adequate clinical and imaging descriptions were included in the study [Table 1].[9,11-13,15,17-20,22,23,25,26] IPC formation

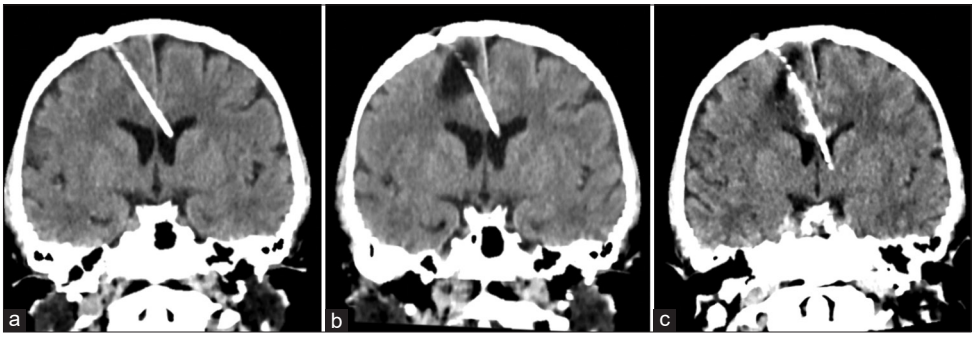

Figure 2: Imaging comparison. CT after VPS surgery: (a) coronal view demonstrating the catheter pathway without any parenchymal lesion. CT at revaluation (3 months later): (b) coronal view demonstrating the catheter pathway with an intraparenchymal pericatheter cyst and perilesional edema, without hydrocephalus. CT after shunt revision (postoperative): (c) coronal view demonstrating the new catheter pathway with cyst regression and mild intracystic hemorrhage.

Table 1: Analysis of "IPC in VPS for hydrocephalus or IIH" bibliography.

Variable	Hydrocephalus	IIH	Overall
No. of cases	32	5	37
Sex	18M: 14F	1M: 4F	19M: 18F
Average age	~17.7 yrs [2 mos–65 yrs]	~32 yrs [22–46 yrs]	~18 yrs [2 mos–65 yrs]
Concomitant/ previous condition	NTDs: ~28.1% (9/32)/SAH or IVH: ~12.5% (4/32)	Chiari I: 20% (1/5)/leukemia: 20% (1/5)	NTDs: ~24.3% (9/37)/SAH or IVH: ~13.5% (5/37)
Clinical presentation at revaluation	Headache~31.2% (10/32)/ vomiting~31.2% (10/32)/motor impairment~28.1% (9/32)/visual changes~6.2% (2/32)	Headache 80% (4/5)/visual changes 40% (2/5)	Headache~40% (14/37)/ vomiting~27% (10/37)/motor impairment~25% (9/37)/visual changes~10.8% (11/37)
Interval from VPS surgery	~3.5 yrs	~1.5 yrs	~3.2 yrs
Shunt revision	Yes: 26/No: 3/NA: 3	Yes: 4/No: 1	Yes: 30/No: 4/NA: 3

IIH: Intracranial idiopathic hypertension, NTDs: Neural tube defects, SAH: Subarachnoid hemorrhage, IVH: Intraventricular hemorrhage, IPC: Intraparenchymal pericatheter cyst, VPS: Ventriculoperitoneal shunt, mos: Months, yrs: Years

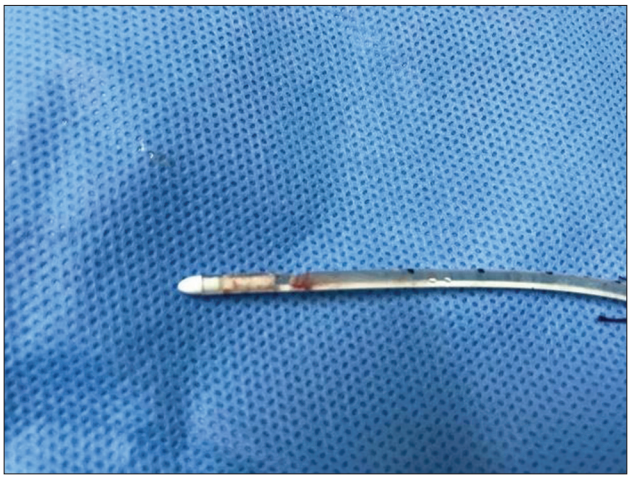

Figure 3: Intraoperative image: view of the obstruction of the proximal catheter tip during shunt revision.

probably results from a weakening of ependymal cells, which, associated with VPS malfunction, cause increased CSF pressure and subsequent parenchymal collection.[21] Then, depending on the degree of the pericatheter gliosis, a cyst can be formed, sometimes associated with vasogenic edema.[8,14] Possibly, another factor is the more compliance of the white matter surrounding the catheter compared to the ependyma. The CSF diffuses through the catheter orifice created in the ependyma and accumulates in the white matter during the course of the occluded catheter. Moreover, those who have IIH without ventricular dilation may present less compliant ventricles. The intracranial pressure increases, but as there is a low resistance outlet (catheter orifice), the liquid exits there, without ventricular dilation. Therefore, the presence of ventricular dilatation may not be a precise or the sole factor to estimate the correct functioning of the shunt system. These mechanisms may explain why such cysts are more common in children, since their cerebral parenchyma and ventricles are softer and more elastic, respectively.[4,14] Children account for 60% of the 35 reported cases.

Patients with this complication present with varied symptoms, the most frequent being headache (~40%), vomiting (~27%), and visual changes (~10%). There have also been reports of decreased levels of consciousness, hemiparesis, speech and gait disorders, tinnitus, and epileptic seizures. One patient was asymptomatic at re-evaluation, and the IPC was found by routine imaging tests.[24] Interestingly, the diagnosis of this complication can occur in a variable time from shunt implant, with cyst onset being reported years, months, and even days after surgery.

Imaging tests are of utmost importance to make a differential diagnosis of parenchymal lesions in VPS patients as well as to identify concomitant conditions significant to the therapeutic approach. The CT examination can assist in the diagnosis of IPCs, and it is very often the initial imaging test. However, MRI is the most helpful examination as it allows the correct distinction between cysts and their differential diagnoses due to its better resolution.[6] Alternative imaging techniques, such as CT cisternography and nuclear medicine scans, can be useful to recognize a cyst-ventricle communication or catheter obstruction, respectively.[4,8]

CSF edema, tumors, abscesses, and dysembryogenetic lesions are all considered cyst-like lesions.[6,14] Porencephalic cysts (or porencephaly) share the same clinicoradiological features as IPCs, although they can be considered a differential diagnosis[6] and are divided into communicating or noncommunicating, depending on whether they are contiguous with the intraventricular and/or subarachnoid spaces. A cyst-ventricle communication was well described in two cases.[8,10] Certain imaging conditions can present concomitantly to the pericatheter cyst, such as CSF edema and hydrocephalus. Dual cyst presentation is also possible.[5]

Perilesional CSF edema was the only known accompanying condition in the presented case [Figures 1 and 2].

Early recognition and treatment of an IPC can provide complete clinical improvement and cyst regression.[5,8] A shunt revision is the most common approach for IPCs (29 of 33, ~88%), typically in symptomatic cases with catheter obstruction. There were no instances of cyst recurrence. However, a new cyst was formed after a contralateral shunt placement[1] and a pseudocyst identification after an apparent cyst regrowth.[2] Both cases were managed with shunt revision (followed by a cystoperitoneal shunt placement in the first). There have only been four reported cases of IPCs after VPS surgery for IIH, all of them in adults, and a shunt revision was performed in three of them, resulting in near-complete cyst resolution.[4] The age and management features were also similar to the presented case.

Some authors have questioned the necessity of shunt revision, especially in asymptomatic patients. Revision was not performed in four cases, three of them being managed conservatively with clinical and imaging surveillance,[5,24] and one being lost to follow-up.[4] No patient who underwent conservative treatment showed symptoms of shunt malfunction. All remained asymptomatic and two evolved with cyst regression.[16] Sinha *et al*.[16] supported conservative treatment, especially in asymptomatic patients. This type of approach has the advantage of avoiding surgery, though an obstruction, even if partial, may lead to the necessity of shunt revision.[14]

CONCLUSION

Although rare entities, it is important to understand and consider IPCs as complications of VPS surgery, including in adult patients and IIH cases. Shunt revision is the standard approach, though conservative treatment has been performed in specific conditions.

Declaration of the patient consent

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

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Aly B, Hammad W, Elkhouly H. Uncommon complications of ventriculoperitoneal shunt: Experience in 22 cases. Al-Azhar Assiut Med J 2015;13:360-73.
2. Amans MR, Dillon WP. Cerebral parenchymal cyst: A rare complication of ventriculoperitoneal shunt malfunction in an adult. Radiol Case Rep 2015;8(1):784.
3. Balasubramaniam S, Tyagi DK, Sawant HV. Intraparenchymal pericatheter cyst following disconnection of ventriculoperitoneal shunt system. J Postgrad Med 2013;59:232-4.
4. Bianchi F, Frassanito P, Tamburrini G, Caldarelli M, Massimi L. Shunt malfunction mimicking a cystic tumour. Br J Neurosurg 2017;31:484-6.
5. Chiba Y, Takagi H, Nakajima F, Fujii S, Kitahara T, Yagishita S, *et al*. Cerebrospinal fluid edema: A rare complication of shunt operations for hydrocephalus. Report of three cases. J Neurosurg 1982;57:697-700.
6. Iqbal J, Hassounah M, Sheikh B. Intraparenchymal pericatheter cyst. A rare complication of ventriculoperitoneal shunt for hydrocephalus. Br J Neurosurg 2000;14:255-8.
7. Jena RK, Agrawal A, Sandeep Y, Shrikhande NN. Intraparenchymal pericatheter cyst in a patient with cerebral shunt malformation. Narayana Med J 2016;5:145-7.
8. Kale HA, Muthukrishnan A, Hegde SV, Agarwal V. Intracranial perishunt catheter fluid collections with Edema, a sign of shunt malfunction: Correlation of CT/MRI and nuclear medicine findings. AJNR Am J Neuroradiol 2017;38:1754-7.
9. Kalovidouri A, Boto J, Vargas MI. Cerebral CSF cyst as a rare complication of ventriculoperitoneal shunt. J Neuroradiol 2016;43:303-5.
10. Karydakis P, Nikas I, Panagopoulos D, Filippidou M, Sfakianos G, Themistocleous M. Intraparenchymal pericatheter cyst after cerebrospinal fluid shunt: A rare complication with challenging diagnosis case presentation and review of the literature. Asian J Neurosurg 2019;14:581-4.
11. Merkler AE, Ch'ang J, Parker WE, Murthy SB, Kamel H. The rate of complications after ventriculoperitoneal shunt surgery. World Neurosurg 2017;98:654-8.
12. Mollan SP, Davies B, Silver NC, Shaw S, Mallucci CL, Wakerley BR, *et al*. Idiopathic intracranial hypertension: Consensus guidelines on management. J Neurol Neurosurg Psychiatry 2018;89:1088-100.
13. Motomochi M, Nakata K, Shindoh H, Minami S, Fukami M, Ueda K, *et al*. Case of reversible porencephalic cyst during malformation of VP shunts. Nerv Syst Child 1987;12:147-50.
14. Palmieri A, Pasquini U, Menichelli F, Salvolini U. Cerebral damage following ventricular shunt for infantile hydrocephalus evaluated by computed tomography. Neuroradiology 1981;21:33-5.
15. Park HJ, Lee SH, Park HJ, Shin SH. A porencephalic cyst formation in a 6-year-old female with a functioning ventriculoperitoneal shunt: A case-based review. Childs Nerv Syst 2018;34:611-6.
16. Rim HR, Hwang SK, Kwon SH, Kim HM. Intraparenchymal pericatheter cyst as a complication of a ventriculo-peritoneal shunt in a premature infant. J Korean Neurosurg Soc 2011;50:143-6.
17. Sakai N, Arai Y, Hirayama H. Malfuctioning ventricle-peritoneal shunt, associated with porencephaly. Nerv Syst Child 1985;10:427-32.

18. Sakai N, Yamanouchi Y, Numa Y, Matsumura H. Reversible porencephaly associated with shunt malfunction. Nerv Syst Child 1989;14:365-8.
19. Sakamoto H, Fujitani K, Kitano S, Murata K, Hakuba A. Cerebrospinal fluid edema associated with shunt obstruction. J Neurosurg 1994;81:179-83.
20. Santín-Amo JM, Rico-Cotelo M, Serramito-García R, Gelabert-González M, Allut AG. Cavidad porencefálica reversible secundaria a disfunción valvular [Reversible porencephalic cyst related to shunt dysfunction]. Rev Neurol 2014;58:255-8.
21. Shekawat JS, Sundar IV, Poonia N, Sinha VD. Intraparenchymal pericatheter cyst following ventriculoperitoneal shunt. Neurol India 2012;60:341-2.
22. Sinha AK, Lall R, Benson R, O'Brien DF, Buxton N. Intraparenchymal pericatheter cyst following ventriculoperitoneal shunt insertion: does it always merit shunt revision? Zentralbl Neurochir 2008;69:152-4.
23. Sugimoto K, Enomoto T, Nose T. Reversible porencephaly. Alteration of the cerebrospinal fluid flow after shunt malfunction. Childs Nerv Syst 1991;7:394-8.
24. Vajramani GV, Fugleholm K. Reversible CSF cyst related to a functioning ventriculo-peritoneal shunt. Acta Neurochir (Wien) 2005;147:1199-202.
25. Wallace DJ, Grandhi R. Intraparenchymal pericatheter cyst in ventriculoperitoneal shunt failure. BMJ Case Rep 2017;2017:bcr2017220468.
26. Watkins JD, Lee J, Van Engen MJ, Tibbs ML, Ellegala DB, Nicholas JS. Isolated left homonymous hemianopia secondary to a pericatheter cyst-a rare presentation of a ventriculoperitoneal shunt failure. J Neuroophthalmol 2015;35:60-4.

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