CASE REPORT

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FRACTURE INTO THE ABDOMINAL CAVITY AND THE PRESENCE OF A RETROAURICULAR PSEUDOCYST ARE DIAGNOSTIC OF A PARTIAL VENTRICULOPERITONEAL SHUNT DYSFUNCTION

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Abstract. *Ventriculoperitoneal shunt malfunction is frequently observed, with the ventriculoperitoneal shunt fracture being a prevalent etiology. The occurrence of ventriculoperitoneal shunt malfunction is approximately 40% in the first year after implantation and increases to 50% in the second year. Ventriculoperitoneal shunt fracture accounts for 15% of total ventriculoperitoneal shunt malfunction incidents. Ventriculoperitoneal shunt fractures commonly occur due to calcification, immune reactions, and abrasions. The report describes a 5-year-old child who exhibited ventriculoperitoneal shunt malfunction based on clinical examination and CT scan findings. The patient presented with fever and pain in the postoperative wound located posteriorly to the left ear. Radiographic imaging of the cranium and thoracoabdominal region revealed the presence of a ventriculoperitoneal shunt fracture and a C5 spinal disconnection. Additionally, it was demonstrated that the distal catheter had relocated to the abdominal cavity. The laboratory examination and cerebrospinal fluid analysis yielded normal results. The fractured ventriculoperitoneal shunt was subsequently removed and replaced with a new one. It is important to thoroughly investigate and comprehend ventriculoperitoneal shunt malfunctions, even during routine follow-ups.*

Key words: *ventriculoperitoneal shunt malfunction, ventriculoperitoneal shunt fracture, cranium, abdominal cavity, complications*

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INTRODUCTION

Since John Holter's invention of the shunt valve in 1959, ventriculoperitoneal shunt (VPS) has been the primary treatment technique for hydrocephalus. Although complications have been reduced, the therapy of this frequent neurosurgical illness is still difficult [1].

During the first year following implantation, the incidence of VPS malfunction is roughly 40%, and in the following year, that number rises to 50%. The failure of a VPS can be brought on by a number of factors, including infection, excessive drainage, and a variety of mechanical issues, including obstruction, migration, disconnection, and fracture. Mechanical VPS failure might be difficult to diagnose because of

conflicting reports in the medical literature on the effects of disconnected or broken shunts. Concerning patient follow-up, diagnostic procedures, and treatment strategy, a wide range of perspectives has been recorded [2].

One type of VPS dysfunction is a fracture. 15% of all incidents of VPS malfunction involve a fractured VPS. Before the ages of 15 and 17, VPS fractures are more common in young boys than in adults. Immune responses, calcification, and abrasion can result in VPS dysfunction and cause VPS fracture. Repetitive twisting movements of the neck and persistent shunt stretching, when fibrosis from the development spurt has bound it, together are two risk factors that contribute to VPS fracture. In some cases of VPS fracture, cerebrospinal fluid (CSF) flow still occurs, even when there is no good drainage because of the skin tunnel formed by tissues around the catheter [2, 3].

In pediatric scenarios, VPS fractures are most frequently seen throughout the patient growth phase and as their height increases. The connection side of the repetitive pressure is where the majority of VPS fracture cases are reported. Fractures typically occur in the lateral-cervical region, the temporal site, or the chest wall region, all of which correspond to prevalent calcification sites and stretching catheter zones [2, 3].

CASE PRESENTATION

A 5-year-old child was referred from a peripheral hospital due to pain in the surgical wound behind their left ear for the past week before admission. The pain was intermittent and accompanied by swelling increasing in size around the left Keen's point where the VPS was inserted. There were no symptoms of high intracranial pressure.

The patient relatives reported three-year seizures based on the prior hospital medical record and VP shut inserted in the same year. During the seizure, the patient's right hand and right leg began to twitch in unison, while the patient was running a fever. The patient was referred to our hospital and was diagnosed with Dandy–Walker syndrome [4, 5] porencephalic cyst, and hydrocephalus. The patient had a left VPS insertion on Keen's point, and the surgery was performed on May 13, 2016. The patient did not visit routinely the Neurosurgery or Pediatric Clinic for follow-ups.

Based on the clinical examination, swelling and pain were found in the retroauricular region (Fig. 1A,B,C). There was no hyperemia in this area or on the peritoneal catheter tract. The score on the Glasgow Coma Scale (GCS) was 15. The pupils were the same size and moved in response to the light. There were no signs of meningitis.

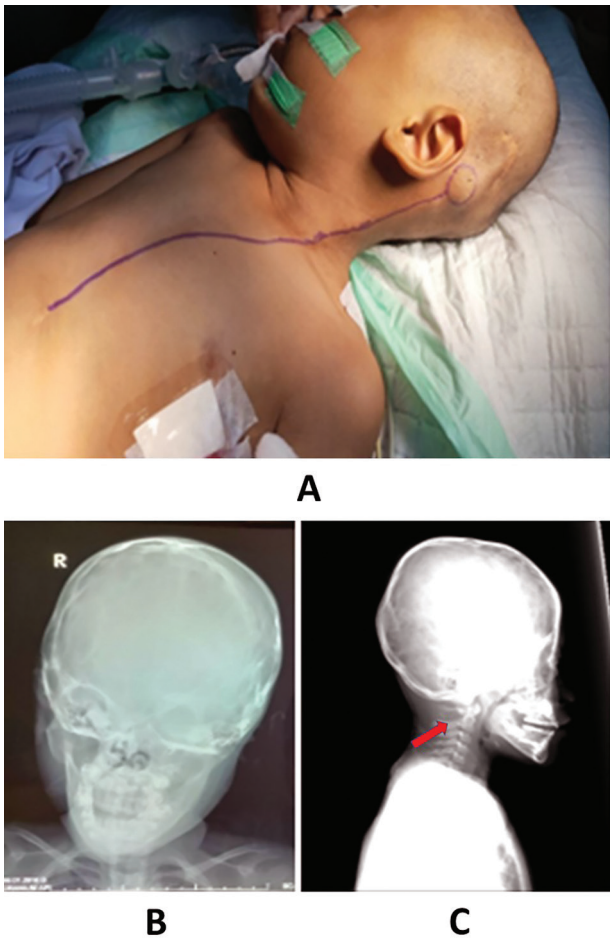

Fig 1. A: Swelling around the chamber of the VPS on the retroauricular region, B–C: Skull radiography AP/Lat., respectively

The skull and thoracoabdominal radiography on July 28, 2021, showed a shunt fracture and disconnection at the level of the fifth cervical spine (Fig. 2) and a free distal catheter in the abdominal cavity (Fig. 2).

The patient's laboratory examination and CSF analysis were normal. A computed tomography (CT) scan with contrast on July 28, 2021, revealed no active hydrocephalus and no signs of meningitis, but Dandy–Walker syndrome, porencephalic cyst, and cerebellar edema were still present (Fig. 3A).

Based on the findings, the patient was diagnosed with a partially malfunctioning ventriculoperitoneal shunt due to a shunt fracture into the abdomen, with retroauricular pseudocyst. During the operation, we found a fracture of the catheter at the fourth cervical spine level. The distal part of the peritoneal catheter migrated into the peritoneal cavity, and therefore, shunt removal and reinsertion of the new shunt were

necessary. The CSF was still drained through the tissue tunnel of the peritoneal catheter. On August 4, 2021, removal of the fractured shunt and reinsertion of the replacement shunt at the left Kocher's site were accomplished. Brain swelling subsided, yet hydrocephalus is still active, as seen on a CT scan (Fig. 3B) taken after surgery.

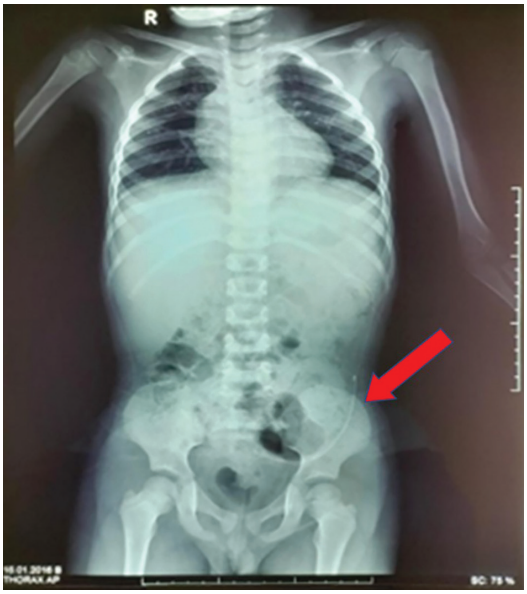

Fig. 2. Thoracoabdominal radiograph presenting a catheter placed distally in the abdominal cavity

DISCUSSION

The challenges associated with VPS can be broken down into two categories: mechanical and non-mechanical. Some of the mechanical issues include blockage, disconnection, and movement. Infection and distal compartment-related issues such as pseudocyst development, ascites, and pleural effusion are examples of the non-mechanical consequences [6]. A lack of regular epidemiological data has hindered disease awareness and caused inequitable resource allocation for patient treatment and research [7]. In this study, misplaced hardware and proximal occlusion were the most common complications in children. 30-day complication rates imply a high rate of additional problems. Multi-targeted techniques that reduce misplacement may lower revision rates [8].

Shunt migration is the relocation of a portion or the entire shunt system (proximal or distal catheter/reservoir/valve) from its designated compartment to a new compartment, which may or may not be related with shunt failures [6]. In this case report, the distal catheter of the VPS was found migrating to the peritoneal cavity with a partial malfunction of the VPS.

Subcutaneous pseudocyst usually presents as a localized mass. Pseudocyst of the retroauricular region is a benign cyst resulting from the accumulation of the CSF in the subcutaneous space because of a disconnection of the shunt tube [6]. In this case, based on clinical and laboratory examinations, there was no evidence of infection.

The most typical signs of shunt malfunction include headaches, nausea, and sleepiness, yet the patient in this case did not report any of those symptoms. This is because there was still CSF drainage through the pseudotubing around the space of the catheter, albeit not too adequate. Therefore, not all cases of shunt fracture are related to malfunction; some cases show only partial malfunction. The suggested methodology is a realistic, easy, and minimally invasive

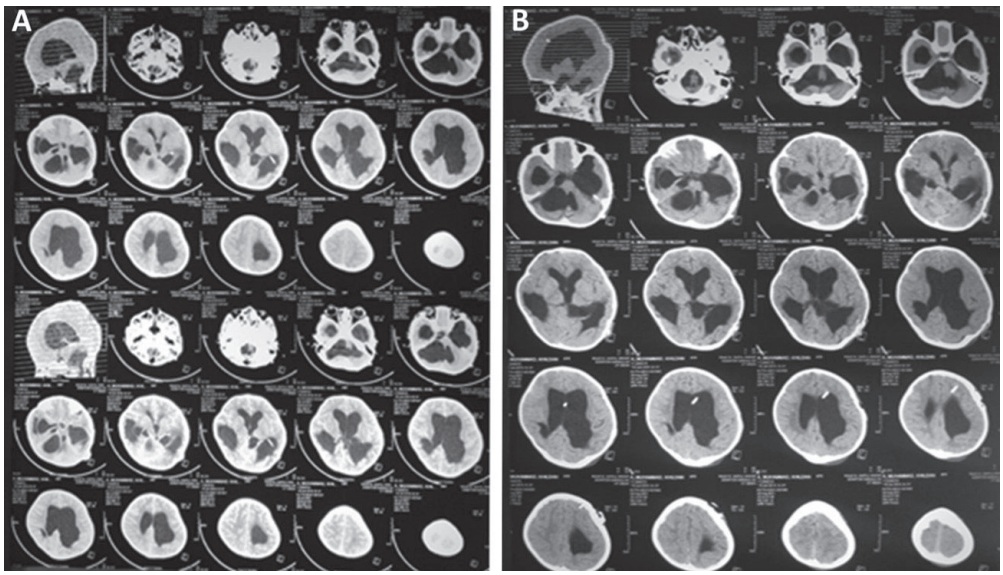

Fig 3. Preoperative (A) and Postoperative CT scan (B)

method for properly diagnosing VPS malfunction, detecting the exact level of system failure, and tailoring surgical VPS repair to avoid unnecessary system replacement [9].

Patients having inserted shunts must be monitored throughout time in order to detect shunt malfunctions in a timely manner and to identify circumstances that provide a risk for shunt fractures. In addition to clinical follow-ups, routine radiological follow-ups, such as brain CT scans or MRIs, are frequently conducted [10].

Some fractures can only be accidently located during normal scar [4, 11]. By functioning as a channel, scar tissue around the shunt catheter may allow cerebrospinal fluid (CSF) to travel between the catheter's two broken ends, as seen in this youngster, whose auricle protruded around the catheter [12].

There are instances where a severed distal segment may enter the peritoneal cavity, but unless there are symptoms of an infection in the abdomen or unexplained abdominal pain, this condition can proceed unnoticed. Before deciding to remove a fractured catheter segment, an abdominal examination is necessary [11]. Our patient was found to have bifurcation fractures of the skull and abdomen. There were no abdominal symptoms. Therefore, the removal of the fractured catheter shunt was not necessary [13, 14, 15].

Shunt disconnection and fracture accounted for 15% of shunt failures in a study of Aldrich and Harmann, and occipitally positioned shunts were more likely to dislocate than those at the front of the body [16]. Unishunt systems effectively address and resolve this particular concern where multicomponent systems are sometimes necessary, and many surgeons choose to link the ventricular catheter to a proximal valve or reservoir. VP shunts with ventricular catheter-reservoir connections in the frontal area, where motion is limited, are not likely to detach. Avoid occipitally implanted shunts with distal connections [16]. This research confirms previous findings that 6.66% of occipitally-placed shunt problems involve shunt fracture. In addition, it was discovered that occipitoparietal VP shunts are more prone to problems than frontal VP shunts. Specifically, 10.4% of patients undergoing occipitoparietal VP shunt and 6% of those undergoing frontal type experienced problems [16].

Previous research has shown that both age and etiology play a role in determining how likely it is that a shunt would break first [17, 18, 19]. In line with our findings, recent research has shown that age and etiology may be highly related to shunt revision for a variety of reasons, including growth and bone development.

Detailed clinical tests and imaging, such as radiography and CT scans, should be undertaken when a patient comes with pseudocyst and symptoms of elevated ICP to detect the risk of shunt fractures.

CONCLUSION

Shunt fracture may cause a total or partial malfunction of the shunt. A pseudocyst can cause a partial malfunction of the shunt due to the accumulation of CSF in the subcutaneous space. Pseudocyst on the shunt tract should be investigated if there is shunt fracture or not. Additionally, CSF drainage can be evaluated clinically and by CT re-evaluation.

In this case, treating the patient with VPS fracture accompanied by retroauricular pseudocyst and free distal catheter in the abdominal cavity without any infection only required the removal of the shunt, replacing it with a new one and leaving a free distal catheter in the abdominal cavity.

Informed Consent Statement: *This study has gained Informed consent from the patient family.*

Disclosure Summary: *The authors have nothing to disclose.*

REFERENCES

1. Park MK, Kim M, Park KS, et al. A Retrospective Analysis of Ventriculoperitoneal Shunt Revision Cases of a Single Institute. J Korean Neurosurg Soc, May 2015, 57(5), 359-363. doi: 10.3340/jkns.2015.57.5.359.
2. Erol FS, Ozturk S, Akgun B, Kaplan M. Ventriculoperitoneal shunt malfunction caused by fractures and disconnections over 10 years of follow-up. Childs Nerv Syst, Mar. 2017, 33(3), 475-481. doi: 10.1007/s00381-017-3342-0.
3. Jibia A, Oumarou BN, Adoum M, et al. Repeat fracture of shunts in ventriculoperitoneal shunting with pelvic migration: An African teen case report with literature review. Interdiscip. Neurosurg, 2022, 27(101381). doi: 10.1016/j.inat.2021.101381.
4. Jadhav SS, Dhok A, Mitra K, et al. Dandy-Walker Malformation With Hydrocephalus: Diagnosis and Its Treatment. Cureus, May 2022, doi: 10.7759/cureus.25287.
5. Mohanty A, Biswas A, Satish S, et al. Treatment options for Dandy-Walker malformation. J Neurosurg, Nov. 2006, 105(5 Suppl), 348-356. doi: 10.3171/ped.2006.105.5.348.
6. Yim SB, Chung YG, Won YS. Delayed Abdominal Pseudocyst after Ventriculoperitoneal Shunt Surgery: A Case Report. The Nerve, Oct. 2018, 4(2), 111-114. doi: 10.21129/nerve.2018.4.2.111.
7. Isaacs AM, et al. Age-specific global epidemiology of hydrocephalus: Systematic review, metanalysis and global birth surveillance. PloS One, 2018, 13(10), e0204926, doi: 10.1371/journal.pone.0204926.

8. Mansoor N, Solheim O, Fredriksli OA, Gulati S. Shunt complications and revisions in children: A retrospective single institution study. Brain Behav, Nov. 2021, 11(11), e2390. doi: 10.1002/brb3.2390.
9. Broggi M, et al. Diagnosis of Ventriculoperitoneal Shunt Malfunction: A Practical Algorithm. World Neurosurg. May 2020, 137, e479-e486. doi: 10.1016/j.wneu.2020.02.003.
10. Pinto FCG, de Oliveira MF. Laparoscopy for ventriculoperitoneal shunt implantation and revision surgery. World J Gastrointest Endosc, 2014, 6(9), 415-418. doi: 10.4253/wjge.v6.i9.415.
11. Kang JK, Lee IW. Long-term follow-up of shunting therapy. Childs Nerv Syst, Nov. 1999, 15(11-12), 711-717. doi: 10.1007/s003810050460.
12. Kaplan M, Cakin H, Ozdemir N, et al. Is the elapsed time following the placement of a ventriculoperitoneal shunt catheter an individual risk factor for shunt fractures? Pediatr Neurosurg, 2012, 48(6), 348-351, doi: 10.1159/000353616.
13. Bokhari I, Rehman L, Hassan S, Hashim MS. Dandy-Walker Malformation: A Clinical and Surgical Outcome Analysis. J Coll Physicians Surg-Pak JCPSP, 2015, 25(6), 431-433.
14. Hamdan AR. Ventriculoperitoneal shunt complications: a local study at Qena University Hospital: a retrospective study. Egypt J Neurosurg, 2018, 33(1), 8. doi: 10.1186/s41984-018-0008-5.
15. Zahedi S, et al.Investigation of ventriculoperitoneal shunt disconnection for hydrocephalus treatment. J Neurosurg Pediatr, Feb. 2021, 27(2), 125-130, doi: 10.3171/2020.6.PEDS20454.
16. Aldrich EF, Harmann P. Disconnection as a cause of ventriculoperitoneal shunt malfunction in multicomponent shunt systems. Pediatr Neurosurg, 1991 1990, 16(6), 309-311; discussion 312. doi: 10.1159/000120549.
17. de Oliveira R S, Barbosa A, de M Y. A. et al. An alternative approach for management of abdominal cerebrospinal fluid pseudocysts in children. Childs Nerv. Syst. ChNS Off. J Int Soc Pediatr Neurosurg, 2007, 23(1), 85-90, doi: 10.1007/s00381-006-0183-7.
18. Patwardhan RV, Nanda A. Implanted ventricular shunts in the United States: the billion-dollar-a-year cost of hydrocephalus treatment. Neurosurgery, 2005, 56(1), 139-144; discussion 144-145, doi: 10.1227/01.neu.0000146206.40375.41.
19. Tuli S, Drake J, Lawless J, et al. Risk factors for repeated cerebrospinal shunt failures in pediatric patients with hydrocephalus. J Neurosurg, 2000, 92(1), 31-38. doi: 10.3171/jns.2000.92.1.0031.