Global Pediatric Health
Volume 11: 1–5
© The Author(s) 2024
Article reuse guidelines:
sagepub.com/journals-permissions
DOI: 10.1177/2333794X241291750
journals.sagepub.com/home/gph
Sage

Trans-Oral Protrusion of the Distal End of a Ventriculoperitoneal Shunt: A Case Report of an Unusual Complication

Joseph Y. B. Nnang, MD[1,2*], Berjo Takoutsing, MD[1*], Lisa Akob, MD[1,3], Gilbert Yada, MD[1,4], Geneviève Endalle, MD[1,5], Mohammed Njoya, MD[1,3], Yvanah Owoundi, MD[1], Astel Dongmo, MD[1,4], Mercy Berinyuy, MD[1,4], Wunde Njineck, MD[1,3], Mustafa Semougnal, MD[1,4], Megan E. H. Still, MD[1,6], and Ignatius Esene, MD, MSc, PhD, MPH[1,3]

Abstract

Ventriculoperitoneal shunting (VPS) is the surgical technique of choice to manage pediatric hydrocephalus. Despite having good results, it is prone to complications, some of which are rare. This is the case report of a 2-year-old male, with an uncomplicated VPS done at 6 months of age, presenting with vomiting, irritability, anorexia, and drooling. There was an oral protrusion of a tube dripping clear fluid. Imaging studies demonstrated evidence of gastric perforation with a cephalic migration and transoral protrusion of the distal end of the shunt tubing. A gastroplasty, and immediate revision of the distal shunt were done free of any complications. This case report underlines the importance of recognizing and managing trans-oral protrusion of the distal end of VPS system in a timely manner, and raises awareness of this uncommon complication and its potential influence on patient health and survival, given ventriculitis' high lethality.

Keywords

bowel perforation, case report, hydrocephalus, oral protrusion, ventriculoperitoneal shunt

Received May 29, 2024. Received revised September 19, 2024. Accepted for publication September 28, 2024.

Introduction

Hydrocephalus is a disorder characterized by the abnormal accumulation of cerebrospinal fluid (CSF) within the brain's ventricular system.[1] Despite ventriculoperitoneal shunting (VPS) being the first-line surgical treatment, it is prone to complications, of which shunt migration is uncommon. Perforation of large bowel/colon, followed by the migration of the distal VPS catheter into the colon with or without extrusion of the distal VPS catheter through the anal canal, is more common among the hollow visceral perforation by the VPS catheter.[2] Still, migration of the distal VPS catheter into the stomach is a rare clinical entity, which is difficult to diagnose especially when not associated with an oral extrusion, with a shunt/central nervous system (CNS) infection being a potentially deadly complication.[3]

We describe the management and outcome of a rare incidence of the trans-oral protrusion of a distal end of a VP shunt catheter managed at a neurosurgical center in Cameroon, a lower-middle income country in Africa according to the World bank.[4] This work has been reported according to the Surgical CAse REport guidelines.[5]

Case Report

This is a 2-year-old male of African ethnicity, who had an uncomplicated VPS at 6 months of age, indicated for

[1]Winners Foundation, Yaounde, Cameroon
[2]University of Yaoundé 1, Yaoundé, Cameroon
[3]University of Bamenda, Bambili, Cameroon
[4]University of Garoua, Garoua, Cameroon
[5]University of Buea, Buea, Cameroon
[6]University of Florida, Gainesville, FL, USA

*Joint first authors.

Corresponding Author:
Berjo Takoutsing, Research Division, Winners Foundation, Yaounde, Cameroon.
Email: takoutsingberjo@gmail.com

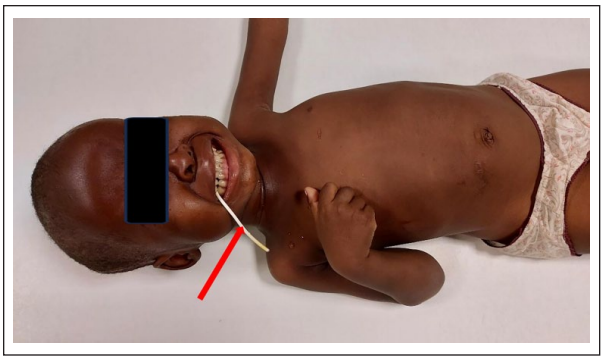

Figure 1. Photo of a 2-year-old child with a trans-oral protrusion of the distal end of a ventriculoperitoneal shunt catheter (Red arrow).

congenital hydrocephalus. He presented to our health facility with persistent vomiting and irritability for 10 days and a refusal to feed for 2 days. This was associated with excessive drooling and the caregivers noticing a tube protruding through his mouth 2 days prior to consultation. The physical examination was notable for a small tube protruding through the patient's mouth in a freely mobile fashion (Figure 1), with signs of moderate dehydration. Signs of meningitis and peritonitis were absent on clinical examination. Head-thoraco-abdominal X-rays were used to evaluate the shunt's integrity and location, and a head-thoraco-abdominal CT scan was done to confirm the diagnosis. There was radiographic evidence of gastric perforation with a cephalic migration and presence within the oral cavity of the distal shunt end (Figures 2 and 3).

The patient was optimized, a pre-anesthesia evaluation was done, and he was taken to the operating room for surgical intervention under general anesthesia. The protruding portion of the distal catheter was carefully resected via the oral cavity. The remaining distal catheter tube was removed via a midline incision (laparotomy). A gastroplasty was done, and a new distal catheter was replaced in the peritoneal cavity (shunt revision). It is worth noting that the tube drained clear CSF, which was sterile on analysis (per- and post-operatively). The proximal end of the shunt was confirmed (clinically by pumping of the shunt valve) to be functioning correctly, and no signs of infection or blockage were noted. Postoperatively, the child was started on intravenous antibiotics (Ceftriaxone: 100 mg/kg body weight 24 hourly for 7 days; Metronidazole: 30 mg/kg body weight for 4 days in 3 divided doses) to prevent any potential infection.

The postoperative period was uneventful, and the patient recovered without complications. The child's oral intake gradually improved, and he was discharged on day 10 post-operatively in stable condition. Follow-up visits were scheduled at 2, 4, 12, and 24 weeks following discharge, the shunt was functioning normally, there was no evidence of infection, and there was a good neurological status. A control thoraco-abdominal X-ray done 2 weeks after shunt revision surgery showed a correctly inserted distal end of the VP shunt into the peritoneal cavity.

Discussion

A migration of the VPS system is one of its relatively uncommonly reported complications.[6] There are 3 types of anatomical migration patterns based on catheter extension and the organs involved: (1) internal, when the catheter invades a natural body cavity; (2) external, when the catheter penetrates the body wall either partially or completely; and (3) compound, when the catheter penetrates a hollow viscus and protrudes through a pre-existing anatomical orifice. In our case, the patient had a compound migration with a gastric perforation and per-oral protrusion of the shunt's distal end, which is relatively rare. The first such case was reported in the literature by Griffith et al in 1987, and several additional cases have been reported.[3] VPS for congenital hydrocephalus, at age $\leq$12 months of age in the male gender is the most common history in affected patients. This patient was a male who had a shunt placed at 6 months old for congenital hydrocephalus. Despite the rarity of this complication, its increasing incidence over the years should prompt clinicians to have a high index of suspicion given its non-specific presentation, especially in cases where there is no obvious oral protrusion. As in this case, most patients present at $\leq$2 years of age and at $\leq$2 years post-VPS.

The stomach is the most common site of perforation in patients with a trans-oral shunt protrusion.[3] This can be attributed to the anatomical proximity of the stomach to the path of the shunt, making it more susceptible to perforation when the shunt migrates into the gastrointestinal tract (GIT).[7] Also, the dynamic nature of the stomach, its exposure to gastric acid, and the pressure exerted by the shunt on the stomach wall contribute to the increased risk of perforation in this organ compared to other parts of the intestine. Oral protrusion of the distal part of the shunt associated with vomiting or mouth drooling is the most common clinical finding observed, as in this case.[3] Also, some may present with GIT symptoms, and a minority with neurological symptoms. In patients with a compound presentation without protrusion of the tube through a natural opening, the presentation is usually non-specific and requires the clinician to have a high index of suspicion.[7] Moreover, fibrosis

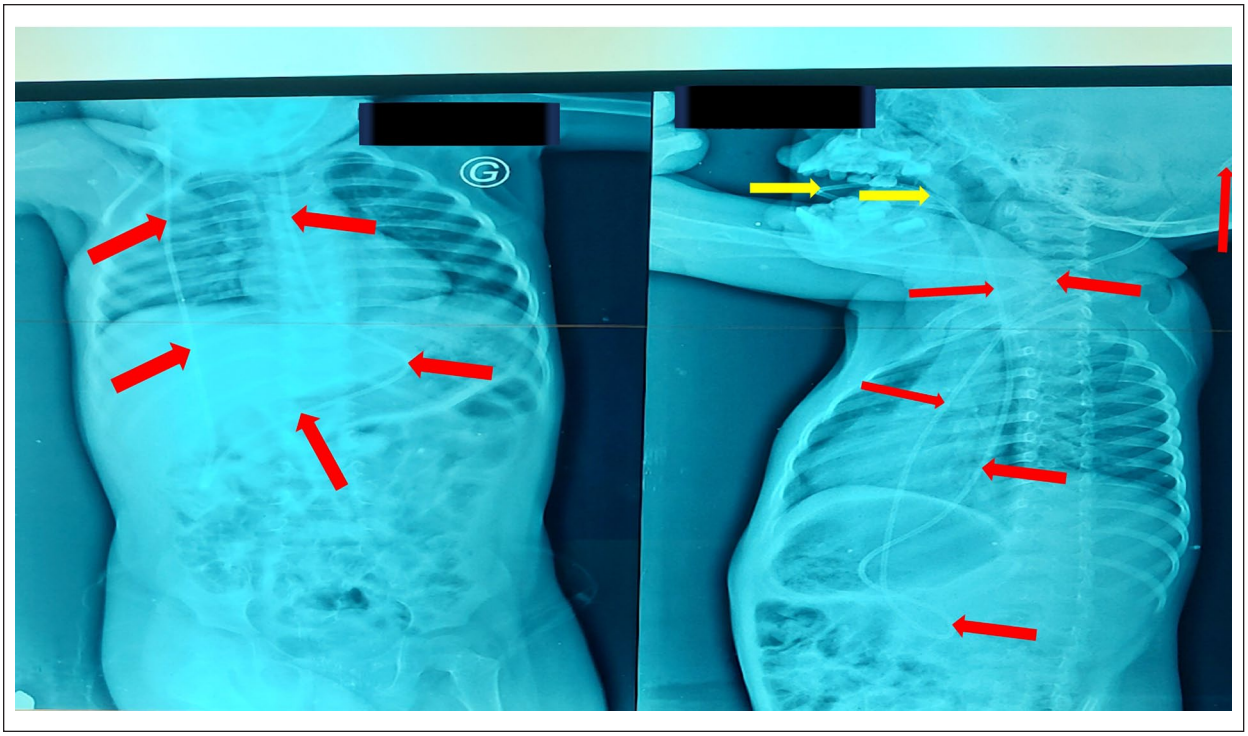

Figure 2. Head-thoraco-abdominal X-rays showing the ventriculoperitoneal shunt system (Red arrows) and oral migration of the distal shunt catheter (Yellow arrows).

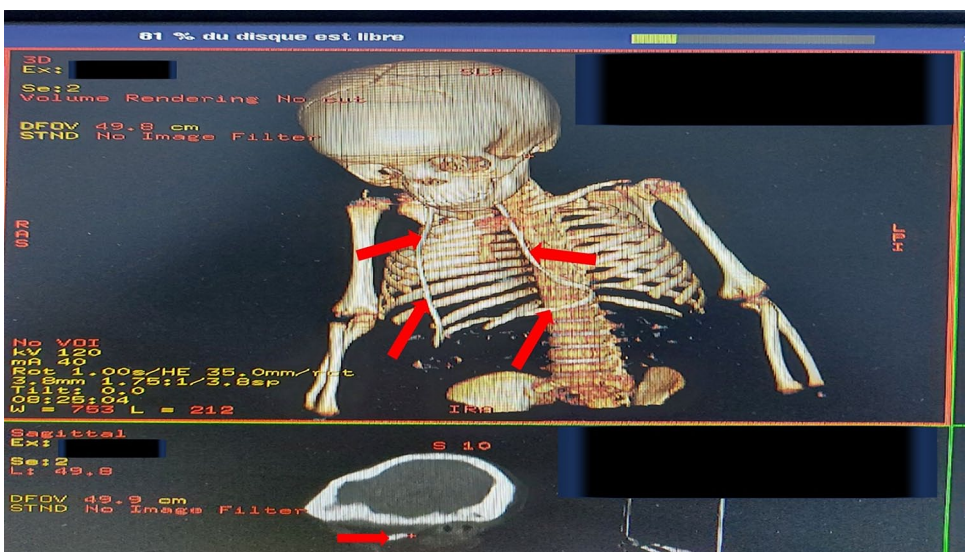

Figure 3. 3-D Head-thoraco-abdominal CT-Scan showing the ventroculoperitoneal shunt system (Red arrows).

around the catheter perforation sites seals the entry into the bowel, preventing peritonitis and thus frequently preventing severe abdominal symptoms, further making it challenging to localize the cause of shunt failure clinically.[3]

Given this context of delayed diagnosis, especially when no extrusion of the shunt is seen, imaging studies are essential in the diagnosis and treatment of shunt complications. It aims to assess the presence of gas under the diaphragm, peritoneal fluid collections, and

the evaluation of the ventricular system. A head-thorax-abdominal X-ray is commonly employed as first-line, according to a recent systematic review by Ghritlaharey.[3] Additionally, a contrast CT scan can detect the existence of shunt/CNS infection, and peritonitis in situations, in addition to aiding in the identification of other shunt complications. In our case, we performed a head-chest-abdominal X-ray, followed by a CT scan, which confirmed the diagnosis and ruled out any other complications. Other investigations include a shuntogram, dye studies, and gastrointestinal endoscopy.

The treatment is surgical, and it is threefold: (1) intragastric migrated distal catheter removal; (2) treatment of the gastric perforation; and (3) with or without shunt revision. Surgical treatment may also differ on a case-by-case basis, depending on the presence or absence of shunt/CNS infection or peritonitis.[8] Nevertheless, the primary treatment goal is to externalize the migrated shunt as soon as possible.[9] Also, gastroplasty may be substituted by fibrin glue application, or in most cases, the gastric perforation is allowed to heal spontaneously.[3] Despite the shunt revision proceeding with the change of the entire shunt system, in our case, only the distal part of the shunt was changed. Also, in a recent systematic review, delayed re-VPS insertion was preferred over immediate shunt revision to have time to treat an associated shunt infection and evaluate the need for maintenance of a VP shunt.[3] In our case, a shunt revision was performed immediately after removing the distal VPS catheter. This decision was guided by the absence of any clinical or laboratory findings suggestive of an infection. The distal shunt catheter drained clear CSF, which was sterile on analysis (per- and post-operatively). In the management of distal shunt migration, intraventricular antibiotic administration is known to shorten the duration of CSF sterilization, the duration of antibiotic use, and the duration of hospital stay.[10] Nevertheless, this is indicated if there is a shunt infection. In this case, there was no evidence of CSF infection, and a systemic antibiotic course was administered with a good outcome.

Conclusion

Trans-oral protrusion of the distal end of a VP shunt following gastric perforation is an uncommon but serious complication. Prompt recognition and appropriate management are essential to improve patient outcomes. This case report highlights the need to consider trans-oral protrusion as a differential diagnosis in pediatric patients with VP shunts presenting with symptoms such as vomiting, drooling, and poor oral intake. Surgical intervention remains the mainstay of treatment, with the goal of removing the protruding distal end of the shunt and closing the perforated orifice, guided by the presence or absence of an associated infection.

Acknowledgments

None declared.

Author Contributions

JYBN, BT, and IE Contributed to the conception, design, drafting, and interpretation of the manuscript. All authors critically revised the manuscript and gave final approval. IE supervised the writing process and is the guarantor. JYBN, and BT contributed equally and are joint first authors.

Declaration of Conflicting Interests

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

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethical Approval

Our institution does not require ethical approval for reporting individual cases.

Consent

Informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

ORCID iD

Berjo Takoutsing https://orcid.org/0000-0002-8993-2060

References

1. Hochstetler A, Raskin J, Blazer-Yost BL. Hydrocephalus: historical analysis and considerations for treatment. *Eur J Med Res*. 2022;27(1):168. doi:10.1186/s40001-022-00798-6
2. Ghritlaharey RK. Systematic literature review of the management of transanal extrusion of distal ventriculoperitoneal shunt catheter: 1966–2020. *Med J DY Patil Vidyapeeth*. 2022;15(5):629-659. doi:10.4103/mjdrdypu.mjdrdypu_149_21
3. Ghritlaharey RK. Migration of the distal ventriculoperitoneal shunt catheter into the stomach with or without trans-oral extrusion: A systematic literature review and meta-analysis. *World J Clin Pediatr*. 2023;12(5):331-349. doi:10.5409/wjcp.v12.i5.331
4. World Bank. *Overview*. World Bank. Accessed March 31, 2024. https://www.worldbank.org/en/country/cameroon/overview
5. Agha RA, Franchi T, Sohrabi C, Mathew G, Kerwan A, SCARE Group. The scare 2020 guideline: updating consensus surgical case report (Scare) guidelines. *Int J Surg*. 2020;84:226-230. doi:10.1016/j.ijsu.2020.10.034

6. Allouh MZ, Al Barbarawi MM, Hiasat MH, Abuzayed BA. Migration of the distal catheter of the ventriculoperitoneal shunt in hydrocephalus patients. *Neurosci*. 2017;22(4):298-302. doi:10.17712/nsj.2017.4.20170137
7. Hai A, Rab AZ, Ghani I, Huda MF, Quadir AQ. Perforation into gut by ventriculoperitoneal shunts: A report of two cases and review of the literature. *J Indian Assoc Pediatr Surg*. 2011;16(1):31-33. doi:10.4103/0971-9261.74521
8. Low SW, Sein L, Yeo TT, Chou N. Migration of the abdominal catheter of a ventriculoperitoneal shunt into the mouth: a rare presentation. *Malays J Med Sci*. 2010;17(3):64-67. Accessed March 31, 2024. https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3216166/
9. Tamlikha A, Shukri F, Zahari Z. Transoral migration of ventriculoperitoneal shunt: a rare presentation. *Gazi Med J*. 2021;32(1):131-134. Accessed March 31, 2024. https://medicaljournal.gazi.edu.tr/index.php/GMJ/article/view/2470
10. Sahin A, Dalgic N, Kilic M, et al. The role of intraventricular antibiotic therapy in the treatment of ventriculoperitoneal shunt infection in children. *Childs Nerv Syst*. 2021;37(5):1605-1612. doi:10.1007/s00381-021-05116-9