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CASE REPORT

Migration of the anal distal end due to ventriculoperitoneal shunt placement: an atypical case report of a 9-month-old infant with tuberculous meningitis and review of the literature

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Abstract

Background Ventriculoperitoneal shunt (VPS) represents one of the most classic and widely used treatments for hydrocephalus in pediatric patients. Migration and externalization of the distal end of the catheter through the rectum are extremely rare complications of intestinal perforation with devastating consequences such as meningitis or peritonitis due to enteric bacteria that are significantly life-threatening. Besides, one of the biggest topics with that is that it can happen without producing symptoms, like the patient we present in this case report, which further masks the condition and puts the patient's life more at risk.

Case presentation We present a case of a 9-month-old infant patient, with a history of prematurity, tuberculous meningitis (TBM), and hydrocephalus, who came to ED with a functional VPS and the distal end of the catheter protruding outside the rectum for 7 days, without presenting neurological or intestinal symptoms accompanying. One of the parameters that guided the diagnosis and made us suspicious of asymptomatic intestinal perforation (IP) was the background of TMB. The patient was immediately transferred to the OR where both ends of the shunt were removed: in the first instance, the shunt tube was disconnected through the abdomen, thus withdrawing through the anus, and subsequently, the proximal end of the catheter was exteriorized. In turn, the intestinal fistula was successfully repaired laparoscopically, and prophylactic antibiotic treatment was early administered. On the 6th postop day, a shunt was internalized, and a child was discharged on postop day 15 without complications with alarm guidelines.

Conclusions The authors of this article strongly suggest that (1) anal extrusion of catheters is an uncommon complication but real: for this reason, its development should be considered in all patients with VPS, especially in infants. (2) The patients are often asymptomatic since false tracts can form around the catheter protecting it from spillage, and thus can be removed without complications. (3) Special care should be taken in patients with conditions that increase the risk of developing IP, such as TMB.

Keywords VPS · Distal anal catheter migration · Tuberculous meningitis · Hydrocephalus

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Introduction

The ventriculoperitoneal shunt (VPS) represents one of the most classically employed treatments for hydrocephalus, being universally accepted [1–3]. It consists of creating a communication between the ventricular cavity and the abdominal peritoneum, thus favoring the correct circulation of CSF, with the aim of reducing ICP and improving the patient's neurological function [4, 5].

However, this procedure is associated with a high percentage of neurological and abdominal complications, which can occur at any time during its evolution, either immediately after the procedure or months or even years after its placement, also that its clinical presentation is extremely heterogeneous, being asymptomatic in the vast majority of cases [6, 7]: which is why this procedure represents a real challenge both for the neurosurgeon who performs it and for the patient who undergoes surgery, besides considering that this is a vulnerable patient as they principally usually are infants [8, 9]. It is known that the degree of presenting this complication is very high in the first years and decreases significantly as the patient grows [2, 5, 10]. For what was mentioned above, it is established that every patient who undergoes such a procedure should be closely monitored for an undetermined time [11].

Intestinal perforation (IP) represents one of the rarest and most feared complications as it may not be accompanied by any symptoms and can cause devastating pictures such as sepsis, peritonitis, and meningitis due to the retrograde ascent of intestinal bacteria to the CNS through the catheter [6, 10, 12].

The medical importance of anal extrusion of the distal catheter lies in its potential correlation with an IP condition, which is why this finding is considered a surgical emergency in every patient, regardless of its severity [13].

Case presentation

A 9-month-old male infant presented to the emergency department (ED) with a ventriculoperitoneal shunt (VPS), placed in another hospital, with a catheter protruding from his rectum for the former 7 days (Fig. 1). The patient had been born prematurely at 36 weeks and had a background of tubercular meningitis (TBM) with congenital hydrocephalus at 6 months of age, without presenting other associated craniospinal anomalies. As of this background, he underwent VPS placement and was administered antituberculosis therapy (ATT) in another hospital, continuing with the medication at the time of his admission. At the physical and neurological examination, the patient was lucid (Glasgow score 14/15), with reactive isochoric pupils and non-protruding fontanelle, afebrile hemodynamically stable, with otherwise typical vital signs, without signs of intestinal obstruction, and no sign of peritonism. The patient was hospitalized immediately, and a thoracoabdominal X-ray was performed, which confirmed shunt continuity and migration to the rectum (Fig. 2). Subsequently, the patient was transferred to the OR where it was decided to proceed and was selected surgical treatment based on the background and neurologic condition: with the proper asepsis and antisepsis measures the distal end of the shunt was removed delicately per anus, after disconnecting in the abdomen, and subsequently, the proximal end was exteriorized of the catheter. The small fistula site located in the distal sigmoid colon was repaired through the laparoscopic route, and augmented by the greater Omentum, performed by the gastrointestinal surgeon, without finding peritoneal effusion during the procedure. The immediate postoperative period was very encouraging: had spontaneous diuresis in the first hour, and feeding commenced once the child was awake with flatus evacuation observed in the first 24 h. Bowel movements occurred 48 h without signs of abdominal distension; in addition, perioperative CSF samples were sterile, but the patient was started on prophylactic board spectrum antibiotics, due to the delicate situation of the patient with a severe background. He remained under observation in the pediatric neurosurgery service for 5 days, and on the 6th postop day, a shunt was internalized again, remaining hospitalized for 10 more days. Finally, on postoperative day 15, he was discharged from the hospital, without neurological sequelae and alarm guidelines and care due to family members. The patient was brought to the clinic the following month for follow-up and medical control, without present intestinal obstruction symptoms or neurological symptoms.

Discussion

Although it is well known that VPS placement correlates significantly with subsequent valvular malfunction, the risk of developing IP clinically associated with extrusion of the anal end of the catheter is extremely rare, ranging from less than 0.05 to 1% [14, 15]. Thus, trans-anal catheter protrusion may represent an indirect correlation with IP, being in many cases the only clinical finding at the time of consultation [16].

Among the associated risk factors in this rare complication, one of the most relevant to mention is a history of tuberculous meningitis (TMB); this is a causal factor in the subsequent development of obstructive hydrocephalus: this represents the main complication as well as a critical prognostic factor of this condition [1, 17]. In turn, TBM is significantly associated with a high rate of mortality and disability in infants who do not receive adequate and early

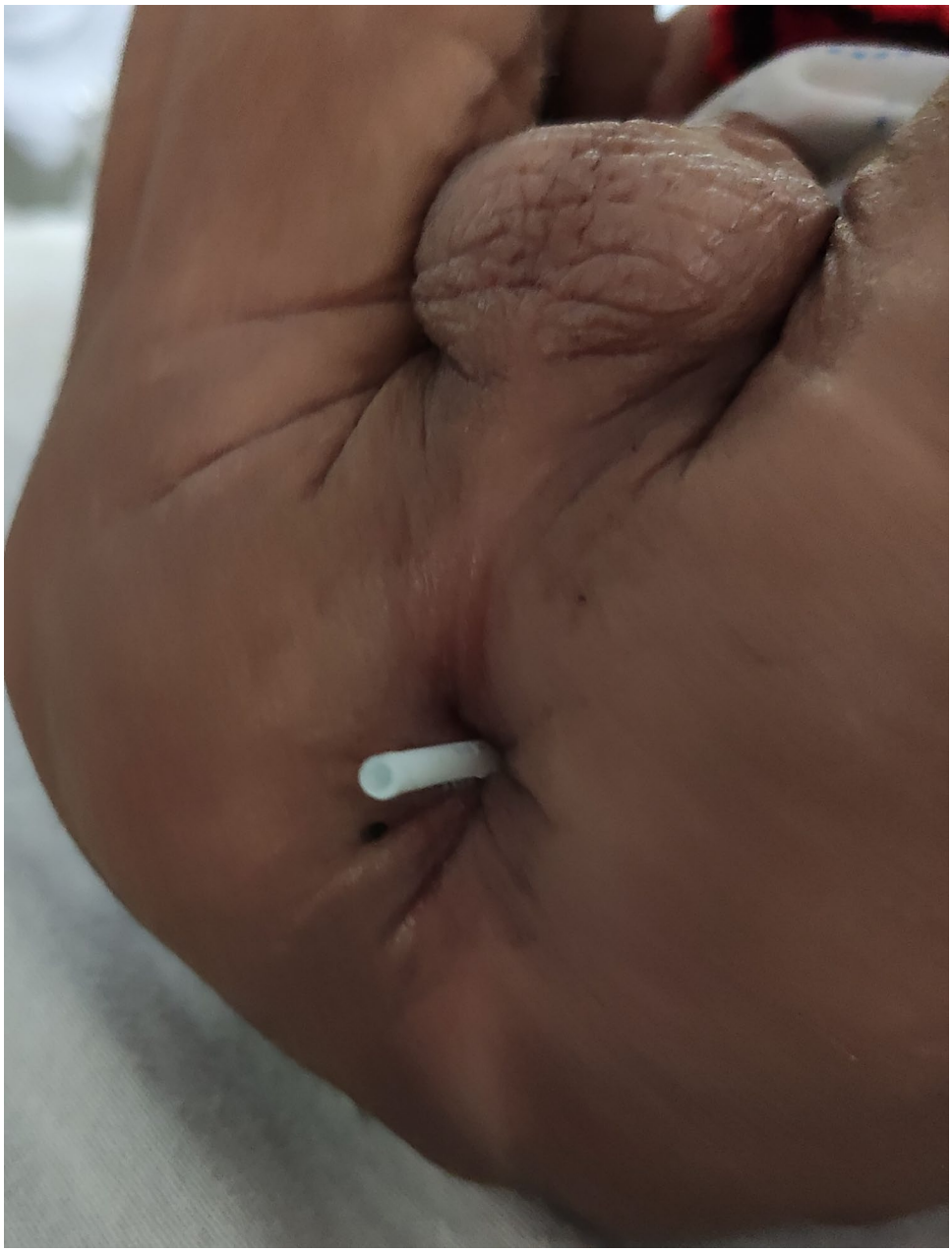

Fig. 1 Clinical photograph showing anal extrusion of VPS

treatment [18]. In addition, other risk factors linked to the development of IP are malnutrition, prolonged corticosteroid treatment, and finally the early age of the patient: because at this age, mainly before the first year of life, there is usually an underdeveloped intestinal wall with high peristalsis [1, 12, 19–21]. Coincidentally, our patient in question meets several of these risk factors, agreeing with the aforementioned literature.

The symptomatology of this condition is extremely heterogeneous since it depends on the complication associated with the catheter extrusion; for this reason, this context represents one of the greatest challenges for the neurosurgeon, given the versatility of the associated symptoms [16]. However, it has been seen that patients who are asymptomatic at the time of the initial consultation usually have a favorable prognosis, in such a way that Sathyanarayana et al. [12] establish that when intestinal perforation does not cause associated symptoms, its prognosis is excellent, as was the case of the patient in the study in question who improved favorably without presenting associated complications.

The pathophysiological mechanism by which catheter migration occurs is currently poorly understood; however, some authors suggest the formation of a fibrous ring around the catheter, thus attaching the catheter to a certain area of the intestinal mucosa, causing no content to enter the peritoneal cavity, thus explaining the lack of symptoms

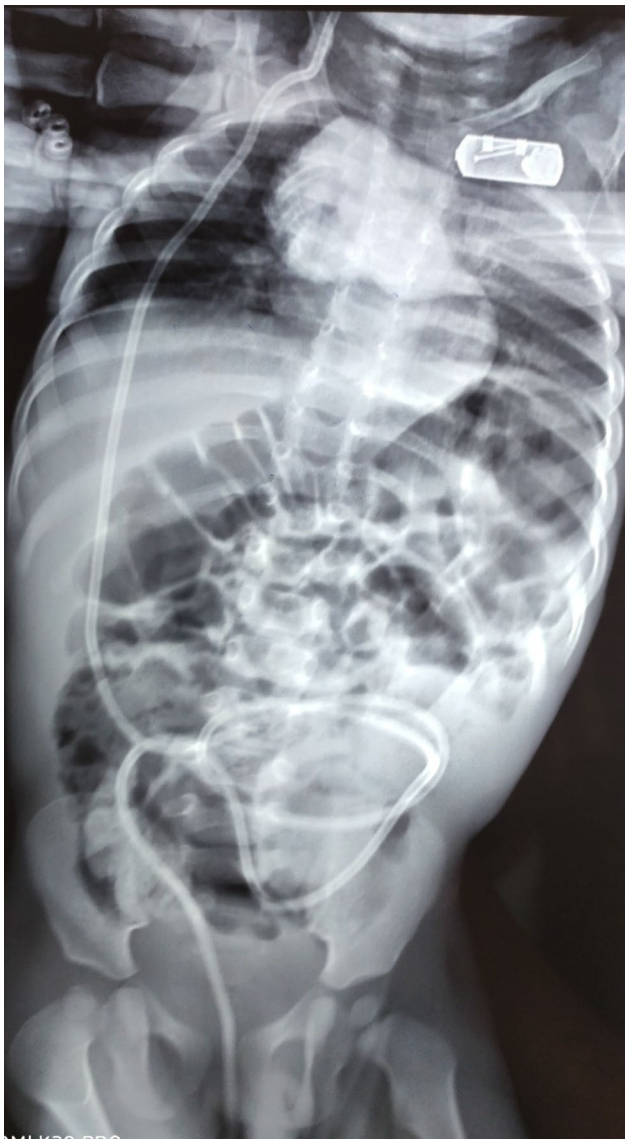

Fig. 2 Thoracoabdominal X-ray confirming the continuity of the VPS and its migration to the rectum

in the patient [22–24]. Thus, Sarkari et al. [11] conclude that more than most cases of VPS-associated bowel perforation occur because of a local inflammatory process and not because of an error in catheter placement. The mortality rate associated with this complication can oscillate around 20–30%, and this can be suggested at the expense of the clinical versatility of this condition which hinders its initial diagnosis, thus generating the subsequent development of potentially fatal complications for the patient such as sepsis, peritonitis or meningitis [25]. On the other hand, the segment of the intestine in which IP occurs is a critical prognostic factor in terms of its evolution, being the large intestine the most frequently affected intestinal portion as well as the most linked to a morbid evolution [26, 27].

The established first-line treatment, if the patient does not present signs suggestive of peritonitis or intestinal obstruction, consists of the removal of the shunt with subsequent intestinal repair using endo-clips and subsequent endoscopic extraction of the catheter with aggressive administration of intravenous antibiotics, also implementing the use of lumbar puncture which should be at hospital admission to rule out the slightest suspicion of meningitis, always observing the patient closely from the time of admission [11, 28, 29]. On the other hand, if there are signs suggestive of peritonitis, obstruction, abscess, or failure to close the fistula after endoscopic extraction, emergency laparotomy is the absolute indication [14].

The objective of Table 1 was to demonstrate the surgical treatment employed in individual case reports versus outcomes in each patient, also taking other variables such as age, gender, primary diagnosis, background, symptoms, CSF culture, and days hospitalized.

We make a comparative table of 22 cases with trans-anal extrusion of the catheter, children under 6 years of age of both sexes published in the literature (2024–2007), in which certain parameters were considered for its selection such as those articles only published in databases: PubMed/MEDLINE, Scopus, and Google Scholar, belonging to recognized Journals, only in the English language.

In this way, we can observe that one of the most common primary diagnoses was congenital hydrocephalus, followed to a lesser extent by postoperative hydrocephalus [22, 32, 43, 45, 46] and communicating hydrocephalus [33, 42]. Also, in the minority of cases, the hydrocephalus following brain trauma or intracerebral hemorrhage has been reported [38]. The background presented by the patients was extremely variable, and the majority were even unknown. Concerning our case report that we present in this article: in the review of the literature, we only found two cases associated with TBM [22, 46], in which a treatment with similar characteristics was performed with favorable outcomes, just like our patient.

As we can see, and this is something that we have detailed previously, this complication decreases in incidence as age increases, with the vast majority of patients being under 36 months (3 years). Another point that we have detailed previously and is notably visible in Table 1: is that most of the patients after presenting said trans-anal catheter extrusion were asymptomatic, their finding often being incidental, just like our patient presented in this case report. On the other hand, it is interesting to mention that the vast majority presented sterile CSF, although in certain cases it was reported CSF: *E. coli*, *Klebsiella*, *P. stuartii*, CoNS, and group B Streptococci [31, 40–42]. However, in all cases, their support was supported with empirical antibiotics, as in our patient in question who always had sterile CSF cultures.

The time to establish the diagnosis after hospitalization was extremely variable: in the vast majority, this information

Table 1 Report of cases in the literature (2024–2007) of patients under 6 years who presented trans-anal extrusion of the distal end of the catheter

AUTHOR	Age	Gender	Primary diagnosis	Background	Symptoms	Time of diagnosis	Image study	Level at which the IP occurred	Surgical treatment	CSF culture	DH (days hospitalized)	Outcomes
Karshe et al. [30]	9 months	M	Cong Hydroc	Cachexia	Asymptomatic	Unknown	X-ray	Ileum	Exploratory laparotomy + total VPS replacement	Sterile	3	No complications
Basehi et al. [31]	24 months	M	Cong Hydroc	Unknown	Asymptomatic	1 h	X-ray	Unknown	VPS exteriorization with a change of both ends + EVD	*P. stuartii*	-	Subdural collection
Khizar and Zahid [32]	42 months	M	Postop Hydroc	MMC	Meningeal signs	10 days	X-ray	Unknown	VPS exteriorization with a change of both ends + fistula repair	Sterile	-	No complications
Heng and Yap [33]	12 months	M	Communic. Hydroc	IVH + Prematurity	Asymptomatic	Unknown	X-ray	Unknown	VPS exteriorization with a change of both ends + fistula repair	Sterile	7	No complications
Hasan et al. [22]	24 months	M	Postop Hydroc	TBM	Asymptomatic	2 days	X-ray	Duodenum	VPS exteriorization with a change of both ends + fistula repair	Sterile	10	No complications
	36 months	M	Cong Hydroc	Unknown	Asymptomatic	1 day	X-ray	Unknown	VPS exteriorization with a change of both ends + fistula repair	Sterile	9	No complications
Indra Gunawan et al. [34]	11 months	F	Cong Hydroc	Unknown	Asymptomatic	Unknown	-	Unknown	VPS exteriorization with a change of both ends + fistula repair	Sterile	-	No complications
Sahoo et al. [35]	24 months	M	Cong Hydroc	Unknown	Asymptomatic	1 day	X-ray	Appendix	Removal of previous VPS + appendectomy	Sterile	-	No complications

Table 1 (continued)

AUTHOR	Age	Gender	Primary diagnosis	Background	Symptoms	Time of diagnosis	Image study	Level at which the IP occurred	Surgical treatment	CSF culture	DH (days hospitalized)	Outcomes
Bodeliwala et al. [36]	10 months	M	Cong Hydroc	Unknown	Asymptomatic	Unknown	X-ray	Appendix	Removal of previous VPS (new contralateral placement) + appendectomy	Sterile	-	No complications
Bansal et al. [37]	12 months	F	Cong Hydroc	DWM + OM	Asymptomatic	Unknown	-	Unknown	Removal of previous VPS + EVD	Sterile	-	No complications
Lee et al. [38]	36 months	M	PTH	TBI + SAH	Abdominal pain	Unknown	X-ray + CT	Ileum	VPS exteriorization with a change of both ends + fistula repair	Sterile	6	No complications
Singh et al. [39]	15 months	M	Cong Hydroc	Unknown	Asymptomatic	15 days	X-ray + USG	Sigmoid colon	VPS exteriorization with a change of both ends + fistula repair	Sterile	7	No complications
Grewal et al. [40]	8 months	M	Cong Hydroc	Unknown	Asymptomatic	Unknown	X-ray	Sigmoid colon	Exploratory laparotomy + total VPS Replacement	Kliebsella	-	No complications
Lawther et al. [41]	5 months	F	Cong Hydroc	MMC + VM	Asymptomatic	Unknown	X-ray	Transverse colon	Distal catheter replacement + primary intestinal repair + EVD	CoNS	21	No complications
Hai et al. [20]	9 months	M	Cong Hydroc	Unknown	Asymptomatic	15 days	Sigmoidesc	Sigmoid colon	VPS exteriorization with a change of both ends + fistula repair	Sterile	-	No complications
	36 months	M	Cong Hydroc	Unknown	Asymptomatic	Unknown	Sigmoidesc	Sigmoid colon	VPS exteriorization with a change of both ends + fistula repair	Sterile	-	No complications

Table 1 (continued)

AUTHOR	Age	Gender	Primary diagnosis	Background	Symptoms	Time of diagnosis	Image study	Level at which the IP occurred	Surgical treatment	CSF culture	DH (days hospitalized)	Outcomes
Chiang et al. [42]	48 months	F	Communic Hydroc	SPM + SMR	Signs of Peritonitis	1 day	X-ray	Sigmoid colon	Exploratory laparotomy + total VPS Replacement	GBS + EC	14	No complications
Vuyyuru et al. [43]	72 months	M	Postop Hydroc	Oligod. Grade III	Asymptomatic	15 days	X-ray + Sigmo	Sigmoid colon	VPS exteriorization with a change of both ends + fistula repair + EVD	Sterile	7	No complications
Jang et al. [44]	36 months	F	Communic Hydroc	BM + BP	Asymptomatic	Unknown	X-ray	Sigmoid colon	Exploratory laparotomy + total VPS Replacement	Sterile	14	No complications
Zhou et al. [45]	8 months	M	Postop Hydroc	MMC	Asymptomatic	Unknown	X-ray	Transverse colon	Exploratory laparotomy + total VPS replacement	Sterile	-	No complications
Handa et al. [46]	18 months	F	Postop Hydroc	TBM	Meningeal signs	Unknown	-	Unknown	VPS exteriorization with a change of both ends + fistula repair	Sterile	-	No complications
	60 months	M	Cong Hydroc	Pelvic abscess	Asymptomatic	Unknown	-	Unknown	Both ends of the catheter were removed and replaced contralaterally.	Sterile	30	Recurrent meningitis + catheter extrusion repeat

Cong Hydroc congenital hydrocephalus, *Communic. Hydroc* communicating hydrocephalus, *PTH* post-traumatic hydrocephalus, *Postop Hydroc* postoperative hydrocephalus, *EVD* external ventricular drainage, *CoNS* coagulase-negative *Staphylococci*, *P. stuartii Providencia stuartii*, *SPM Streptococcus pneumoniae* meningoencephalitis, *EC Escherichia coli*, *GBS* group B Strep, *BM* bacterial meningitis, *BP* bacterial peritonitis, *USG* ultrasonography, *Sigmoidesc* sigmoidoscopy, *IP* intestinal perforation, *MMC* myelomeningocele, *VM* ventriculomegaly, *IVH* intraventricular hemorrhage, *DWM* Dandy-Walker malformation, *SAH* subarachnoid hemorrhage, *TBI* traumatic brain injury, *TBM* tubercular meningitis, *OM* occipital meningocele, *SMR* severe mental retardation, *Oligod.* Oligodendroglioma

was unknown and, in some cases, it could take up to 2 weeks [42, 44]: being 7 days in the case of our patient [33]. The imaging study most used to establish the diagnosis was indisputably the thoracoabdominal X-ray, and in cases minority, it was complemented with other studies such as ultrasound (USG), CT scan, and sigmoidoscopy [20]. In such a way, the most frequently reported site where intestinal perforation (IP), through these studies, occurred was the sigmoid colon [20, 39, 40, 42–44].

The surgical treatment used was extremely versatile, according to the condition and history of each patient: the site of intestinal perforation and the general condition of the patient were taken as individualized parameters. In this way, we can say that in the significant majority, the replacement of both ends of the catheter was used with due asepsis and antisepsis care and the consequent repair of the intestinal fistula, also, in those cases where perforation of the appendix occurred, they were additionally treated by appendectomy [35, 36]. However, in certain cases, the use of external ventricular drainage (EVD) was also implemented [31, 37, 41, 43], as well as in other cases, the repositioning of the catheter on the contralateral side to the original was considered. In those patients with a severity of perforation or a more critical general condition and at the discretion of the neurosurgeon: emergency exploratory laparotomy was considered as treatment [30, 40, 42, 44, 45].

The outcomes in the major number of patients were predominantly favorable without presenting complications with subsequent follow-up after discharge: like our patient in this case report who continues in outpatient observation until now. However, we found that only two patients had poor results: one patient had to undergo a second VPS placement again with several episodes of meningitis [46], and another patient had a small subdural collection after the operation [31].

Conclusions

The authors of this article suggest firmly this rare and severe complication should always be suspected since it can occur in asymptomatic patients, being like the patient in this report, the extrusion of the distal catheter was the only reason for consultation. Besides, timely and early treatment of these rare cases can reduce morbidity and mortality significantly of the infant, as in the case presented in this article which did not present postoperative complications like meningitis, sepsis, or peritonitis due to the retrograde rise of intestinal bacteria. Furthermore, the recent history of tuberculosis infection in SNC, which the infant presented upon his hospital admission, represents a causal agent extremely important with the migration of the distal end of the catheter. We recommend more studies are needed to evaluate this complication linked to VPS as well as to take early measures to prevent it from happening.

Abbreviations VPS: Ventriculoperitoneal shunt; CSF: Cerebrospinal fluid; ICP: Intracranial pressure; IP: Intestinal perforation; TBM: Tuberculous meningitis; ATT: Anti-tubercular therapy; Cong Hydroc: Congenital hydrocephalus; Communic.Hydroc: Communicating hydrocephalus; PTH: Post-traumatic hydrocephalus; Postop Hydroc: Postoperative hydrocephalus; EVD: External ventricular drainage; CoNS: Coagulase-negative *Staphylococci*; *P. stuartii*: *Providencia stuartii*; SPM: *Streptococcus pneumoniae* Meningoencephalitis; EC: *Escherichia coli*; GBS: Group B Strep; BM: Bacterial meningitis; BP: Bacterial peritonitis; USG: Ultrasonography; Sigmoidesc: Sigmoidoscopy; MMC: Myelomeningocele; VM: Ventriculomegaly; IVH: Intraventricular hemorrhage; DWM: Dandy-Walker malformation; SAH: Subarachnoid hemorrhage; TBI: Traumatic brain injury; TBM: Tubercular meningitis; OM: Occipital meningocele; SMR: Severe mental retardation; Oligod.: Oligodendroglioma

Author contribution statement FZ: conceptualization, investigation, methodology, and wrote the main manuscript text, AHS: writing—review editing and prepared Figs. 1 and 2, DNG: investigation and editing Table 1 , LRMS, BC: formal validation. All the authors reviewed the final manuscript.

Availability of data and materials No datasets were generated or analysed during the current study.

Declarations

Ethics approval Ethics approval was conducted and obtained according to the Standards of the Helsinki Declaration; medical research involving human subjects.

Patient consent Obtained.

Conflict of interest There are no conflicts of interest.

Use of artificial intelligence (AI)–assisted technology for manuscript preparation The authors confirm that there was no use of artificial intelligence (AI)–assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

Comments The authors of this article express a rare case report with the purpose of enriching the little published literature on Trans-Anal Migration of the distal end of the Catheter. The authors have Intellectual Property Rights (IPR) over said article in writing, and it can be cited by other authors after its publication in the Journal.

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