



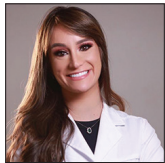
Case Report

Rare case of migration and perforation of the urinary bladder by ventriculoperitoneal shunt catheter with intravesical knotted formation: A case report and literature review

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ABSTRACT

Background: The most commonly used technique in the management of hydrocephalus is a neurosurgical procedure, known as ventriculoperitoneal shunt (VPS). Several complications of the distal end of a VPS catheter have been described in the literature, although migration and erosive bladder perforation of this shunt are extremely uncommon.

Case Description: We relate a rare pediatric case of a 4-year-old boy, with long-term dysuria, intermittent incontinence, and chronic abdominal pain developed during a myelomeningocele postoperative period, without other symptoms. A bladder fistula with the distal end of the VPS inside the bladder was observed while performing an ultrasound to investigate the urinary tract. Two months before the current surgical approach, the patient, with hydrocephalus and alterations compatible with Chiari malformation, was treated with VPS and subsequently subjected to the third ventriculostomy. The child was unsuccessfully treated with an infraumbilical laparotomy, with section and careful traction of the catheter. A vesicostomy was then performed and the catheter was observed to form an intravesical knot.

Conclusion: The intravesical knot formation is extremely rare and its mechanism of formation is still not well understood. Patients with VPS need adequate medical follow-up to identify possible complications of the shunt.

Keywords: Bladder perforation, Intravesical knot, Migration of the distal Ventriculoperitoneal shunt, Urinary and genital tract disorders, Ventriculoperitoneal shunt

INTRODUCTION

Ventriculoperitoneal shunt (VPS) is the technique most commonly used in the management of hydrocephalus, accounting for 0.6% of all pediatric hospitalizations.^[27,28,35] Several complications of the distal end of the VPS have been described in the literature; however, migration and erosive bladder perforation are extremely uncommon.^[26,28,35] We report a rare case of bladder perforation of a 4-year-old boy caused by the distal end of the VPS, with the formation of an intravesical knots, and we perform a literature review.

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METHODS

The study data were obtained retrospectively from the analysis and systematic review of the medical record in question and related literature. The Medline, Central, EMBASE, and Google Scholar databases were used to search keywords such as VPS, VPS, migration of the distal end of VPS, bladder perforation, intracorporeal knotting, and intravesical knotting, in articles published until March 30, 2021. The reference lists generated in the search were then filtered manually for relevance and the reports were selected to remove duplicates. One hundred and ninety-one studies on the migration of the distal end of VPS were found and 44 more relevant bibliographic references were used to write this review. The inclusion criteria for the study were the following: patients who, regardless of age, sex, race, bladder size, and etiology of hydrocephalus, had migration of the distal end of VPS into the bladder, with subsequent extrusion through the urethra or not; patients who had an intracorporeal knot formation at the distal end of the VPS; and patients who had stone formation or calcifications at the distal end of the VPS. The following data were extracted from studies that meet the inclusion criteria: name of the first author, year of publication, age and sex of the patient, clinical and laboratory data presentation, cause of hydrocephalus, size of the urinary bladder, interval since the last insertion of the VPS until revision of the altered VPS, and intervention used to conduct each case and the result.

CASE REPORT

A 4-year-old male patient was admitted to Hospital Infantil Nossa Senhora da Glória – Vitória, ES, Brazil, for the correction of a cerebrospinal fluid fistula and removal of the VPS after evidence of bladder fistula and presence of a catheter inside the bladder. On admission, the child presented increased head circumference, neurogenic bladder, long-term dysuria, constipation, and chronic abdominal pain because of myelomeningocele, but no other associated symptoms. The past pathologic history showed that at 10 months of age, the patient underwent surgery to correct myelomeningocele and for implantation of the right VPS because of hydrocephalus associated with Chiari malformation and that at 3 years of age, after the evidence of ventricular dilatation, the left VPS was inserted. It also showed that after 7 months of this last procedure, the child presented signs and symptoms of meningitis, which grew *Escherichia coli* ESBL in culture of the left VPS catheter tip, being the urine culture also positive for this bacterium. Therefore, he was treated with meropenem, the left VPS catheter was removed and the external ventricular drainage implanted, followed by an endoscopic third ventriculostomy. During an outpatient follow-up with the neurosurgery team, at 4 years of age, asymptomatic, a detailed physical examination was performed, and new

imaging tests were requested, which showed effective control of hydrocephalus. During the same follow-up with the nephrology team, routine ultrasonography of the urinary tract and abdominal X-rays was ordered. A bladder fistula with a VPS catheter in the bladder was found, requiring hospitalization for surgery. In this current hospitalization, the patient was approached by the pediatric surgery and neurosurgery teams through infraumbilical laparotomy with a section of the catheter's distal end and removal of the entire VPS system. After unsuccessfully performing careful tractions to remove the distal portion of the VPS from inside the bladder, they opted for a vesicostomy, at which point was possible to observe the catheter coiled with about eight turns in itself, well intricate and firm, forming an intravesical tangle. No calculus formation was observed and there was no extrusion of the distal catheter through the urethra. He was treated with antibiotic therapy with ceftriaxone and indwelling urinary catheterization for 10 days. The tip of the right VPS catheter was sent for culture, which was negative. The patient follows up on regular outpatient follow-up with neurosurgery and nephrology, progressing uneventfully and without neurological sequelae.

DISCUSSION

The VPS procedure is a well-established treatment modality for hydrocephalus.^[27,28] Migration and perforation of the distal end of the VPS in different regions of the body are relatively uncommon complications.^[3,36,38] Migrations to the pleural cavity, pulmonary artery, breast, heart, navel, vagina, scrotum, bladder, intestine, oral, and anal protrusions have been reported.^[2,4-7,9,12-21,23,25,28-34,37-43]

A case of migration of the distal end of the VPS catheter with intravesical knots formation, without body extrusion, or formation of calcification at the migration site is extremely rare, this being the first report found in the literature. Thirty-one cases in the literature [Table 1], including this one, describe perforation of the urinary bladder by the VPS catheter, 17 of which are related to the extrusion of the distal end of the catheter through the urethra and five of them report the formation of intravesical stones.

Perforation of the urinary bladder through the distal portion of the VPS can be observed in both sexes, with 11 (35.48%) cases being female, 14 (45.16%) males, and six not specified in the reports. It has been reported at all ages, being the youngest case at 8 days old and the oldest at 82 years old; however, it has a higher incidence during childhood due to the VPS procedure being more prevalent at this age.

Some previous reports and discussions^[10,11,22,43] suggest that the occurrence of a VPS perforation may be related to the size of the urinary bladder; however, only 8 (25.80%) patients presented this characteristic in the literature. This case had a

Table 1: Cases reported in the literature on bladder perforation through the distal end of the VPS. [3,5-8,10-12,14,17,18,22-24,26-29,31,33,36,37,39-42]

Author	Year of publication	Number of cases	Age/gender	Urinary bladder size	Neurological disorder (before inserting the shunt)	Approximate duration of the last insertion/revision of the modified VPS	Clinical/laboratory presentation/signs/symptoms
Grosfeld <i>et al.</i> ^[14]	1974	2	3 months/ n.s. 1 year/n.s.	Normal	Hydrocephalus	n.s.	Fever, abdominal pain, and erythema involving lower abdominal wall
Mevorach <i>et al.</i> ^[22]	1992	1	4 years/ female	Increased	Neurogenic bladder and myelodysplasia	n.s.	Extrusion of the distal part of the VPS through the urethra and repetitive UTIs
Prasad <i>et al.</i> ^[29]	1995	1	6 years/ male	Normal	Hydrocephalus and cerebellar medulloblastoma	2 years	Extrusion of the distal part of the VPS through the urethra
Ueda <i>et al.</i> ^[37]	1998	1	82 years/ female	Normal	Hydrocephalus after subarachnoid hemorrhage	10 years	Extrusion of the distal part of the VPS through the urethra and meningitis
Yerkes <i>et al.</i> ^[42]	2001	2	n.s.	Increased in all cases	Neurogenic bladder and myelodysplasia	7 months and 16 years	Meningitis shunt malfunction
Eichel <i>et al.</i> ^[12]	2002	1	3 years/ male	Normal	Posthemorrhagic hydrocephalus	Approximately 3 years	Bladder stone formation, microscopic hematuria, and urinary retention
Surchev <i>et al.</i> ^[36]	2002	1	2 years/ male	Normal	Hydrocephalus secondary to meningoencephalitis	1 year and 7 months	Extrusion of the distal part of the VPS through the urethra
Barker <i>et al.</i> ^[5]	2006	3	6 years/ female 16 years/n.s. 11 years/ male	Increased in all cases	Meningomyelocele and hydrocephalus	5 years 2 years 2.5 years	Fever, abdominal pain, distension, increased serum creatinine, vomiting, and VPS dysfunction
Binning <i>et al.</i> ^[6]	2006	1	14 years/ male	Normal	Congenital hydrocephalus	8 years	Intermittent hematuria
Murthy <i>et al.</i> ^[26]	2009	1	6 years/ male	Normal	Congenital hydrocephalus	4 years	Urinary urgency, dysuria, microscopic hematuria, and bladder stone formation
De Aguiar <i>et al.</i> ^[10]	2011	1	12 years/ female	Increased	Myelomeningocele, hydrocephalus associated with a type II Chiari malformation, and neurogenic bladder	3 months	Lethargy, electrolyte imbalance, and extrusion of the distal part of the VPS through the urethra
Pohlman <i>et al.</i> ^[28]	2011	1	14 years/ male	Normal	Cerebral palsy, delayed development, and hydrocephalus	11 years	Extrusion of the distal part of the VPS through the urethra, small stones in the bladder, and bladder knot
Chen <i>et al.</i> ^[8]	2011	1	Young/ female	Normal	Spontaneous ICH, postoperative hydrocephalus	6 months	Extrusion of the distal part of the VPS through the urethra, hematuria, pyuria, recurrent UTIs, and intra-abdominal abscess

(Contd...)

Table 1: (Continued).

Author	Year of publication	Number of cases	Age/gender	Urinary bladder size	Neurological disorder (before inserting the shunt)	Approximate duration of the last insertion/revision of the modified VPS	Clinical/laboratory presentation/signs/symptoms
De Jong <i>et al.</i> ^[111]	2011	1	38 years	Increased	Hidden spina bifida	n.s.	Extrusion of the distal part of the VPS through an appendicovesicostomy (Mitrofanoff)
Mihajlović <i>et al.</i> ^[23]	2012	1	4 years/ female	Normal	Posthemorrhagic hydrocephalus	4 years	Extrusion of the distal part of the VPS through the urethra
Yazar <i>et al.</i> ^[41]	2012	1	5 years/ female	Normal	Hydrocephalus after posterior fossa medulloblastoma surgery	4 years	Meningitis, recurrent UTIs, and extrusion of the distal part of the VPS through the urethra
Kataria <i>et al.</i> ^[18]	2013	1	7 years/ male	Normal	Congenital hydrocephalus	5 years	Extrusion of the distal part of the VPS through the urethra and intravesical knot
Butler <i>et al.</i> ^[7]	2013	1	9 years/ female	Normal	Congenital hydrocephalus and spina bifida	8 years	Recurrent UTIs, hematuria, abdominal pain, and bladder stone
Ibrahim <i>et al.</i> ^[17]	2014	1	42 years/ female	Normal	Brain tumor	4 years	Dysuria, suprapubic pain, hematuria, urinary urgency, urinary incontinence, recurrent UTIs, and bladder stones
Mutlu <i>et al.</i> ^[27]	2015	1	8 days/ female	Normal	Myelomeningocele and congenital hydrocephalus associated with Chiari malformation type II	1 day	Extrusion of the distal part of the VPS through the urethra
Manuel <i>et al.</i> ^[33]	2016	1	2 years/ male	Normal	Congenital hydrocephalus	1.5 years	Dysuria, urinary incontinence, fever, headache, and extrusion of the distal part of the VPS through the urethra
Miranda <i>et al.</i> ^[24]	2016	1	4 months/ female	Normal	Congenital hydrocephalus	2 months	Irritability, excessive crying, and extrusion of the distal part of the VPS through the urethra
Xu <i>et al.</i> ^[39]	2016	1	38 years/ male	Normal	Hydrocephalus	12 years	Recurrent UTIs, hematuria and urinary incontinence
Al Fauzi <i>et al.</i> ^[3]	2017	1	4 years/ male	Normal	Congenital hydrocephalus and cerebral palsy	2 years	Abdominal pain, extrusion of the distal part of the VPS through the urethra
Rattan <i>et al.</i> ^[31]	2017	1	n.s./male	Normal	Congenital hydrocephalus	10 months	Extrusion of the distal part of the VPS through the urethra
Yang <i>et al.</i> ^[40]	2019	1	29 years/ male	Normal	Post-traumatic hydrocephalus	5 years	Extrusion of the distal part of the VPS through the urethra

(Contd...)

Table 1: (Continued).

Author	Year of publication	Number of cases	Age/gender	Urinary bladder size	Neurological disorder (before inserting the shunt)	Approximate duration of the last insertion/revision of the modified VPS	Clinical/laboratory presentation/signs/symptoms
This case	2021	1	4 years/ male	Normal	Myelomeningocele, congenital hydrocephalus associated with Chiari malformation, and neurogenic bladder	Approximately 3 years	Neurogenic bladder, chronic urinary incontinence, chronic intestinal constipation, chronic abdominal pain, and intravesical knot

VPS: Ventriculoperitoneal shunt, UTIs: Urinary tract infections; n.s.: Not specified

normal sized urinary bladder. The etiology of hydrocephalus was not associated with this complication. The time interval between the last surgery for insertion or reapproach of the VPS and the occurrence of the complication described ranged from 2 months to 12 years.

Five (16.2%) patients had the formation of intravesical stones. Considering that the process and time involved in the formation of a stone are long, it can be speculated that the shunt may remain asymptomatic inside the bladder for a long period before the diagnosis.^[24] Mutlu *et al.* (2015) reported the case of an 8-day-old patient, whose urinary bladder was punctured with extrusion of the distal end of the catheter 1 day after insertion of the VPS. Usually, it is not possible to define precisely the time interval between the bladder perforation by shunt and the diagnosis.

Several theories have been proposed to explain the exact mechanism and facilitators for bladder perforation, such as bioreactivity of catheter components, local inflammatory reaction, infection, relatively rigid and sharp tipped catheters, continuous mechanical irritation at a fixed point on the bladder surface, thin wall of this organ in the pediatric age group, iatrogenic placement with inadvertent positioning of the distal end of the VPS, and long abdominal end of the catheter.^[8,26,28,36,38] However, there is still no unanimous explanation for what happened. The anatomical location of the bladder makes the site highly unlikely to perforate, as the catheter must pass through the peritoneum into the extraperitoneal space and subsequently perforate the bladder, which has a thicker wall and is a relatively more muscular organ than other probable perforations, such as the intestine, for example.^[28,36]

The use of trocars to access the peritoneum for the implantation of the VPS system was associated with an increased risk of perforation of the viscera when compared to open procedures.^[36] However, in our patient, all VPSs performed were through an open procedure and iatrogenic injury to viscera such as the urinary bladder cannot be considered.

The most prevalent clinical signs and symptoms were the extrusion of the distal part of the VPS through the urethra and those related to the urinary tract such as dysuria, urinary urgency, urinary retention, recurrent urinary tract infections, and abdominal pain. Extrusion of the distal portion of the catheter through the urethra was present in 17 (54.83%) patients. Symptoms related to the urinary tract were present in 11 (35.48%) patients; however, as many of them were chronic, a possible overlap of symptoms due to primary neurological disorder (neural tube defects) with symptoms caused by the intravesical catheter should be considered. It is pertinent, therefore, that patients with VPS with urinary symptoms undergo an ultrasound of the urinary and/or abdominal pathways.

Our patient was asymptomatic on admission, with no signs of catheter extrusion through the urethra on physical examination, with the vesical fistula and the presence of the distal end of the VPS inside the bladder only being evident after the abdominal radiography [Figure 1] and urinary tract ultrasonography [Figure 2] routinely requested by the nephrologist, by whom the patient is monitored due to neurogenic bladder and chronic fecal retention resulting from myelomeningocele. Imaging plays a key role in assessing VPS failure and determining the need for surgical revision. An in-depth knowledge of the function and anatomical appearance of the different types of VPS systems, their modes of failures and complications, as well as the corresponding findings in the images are necessary to ensure the timely diagnosis and to prevent permanent neurological damage.^[8,19]

Among the most chosen procedures for surgical approach, there are gentle catheter traction, endoscopic surgery, and exploratory laparotomy.^[1] Most authors choose to remove the catheter mainly through open abdominal surgery.^[8,28,33,41] When there is a periurethral presentation of the catheter, less invasive methods can be chosen, such as pulling the catheter out through the urethral orifice followed by urological

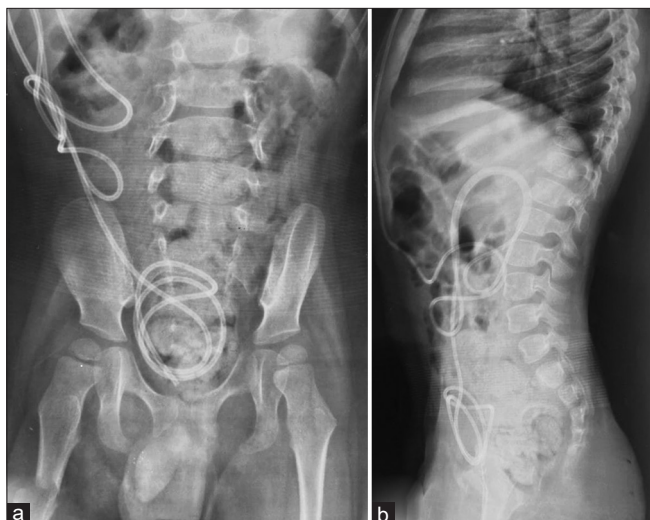


Figure 1: Simple posteroanterior (a) and lateral (b) abdominal radiography demonstrating the presence of a ventriculoperitoneal shunt (radiopaque) catheter in the abdomen, inserting into the urinary bladder.



Figure 2: Ultrasonography of the urinary tract showing bladder fistula with the presence of a ventriculoperitoneal shunt catheter inside the urinary bladder.

endoscopic exploration to visualize the size and location of the perforation and determine the ideal treatment.^[3]

Here, it was decided to perform an exploratory laparotomy followed by successive and careful traction of the catheter that was inserted into the bladder, however, without success. Hence, it was decided to perform a vesicostomy. Finally, when removing the distal end of the catheter from the inside of the urinary bladder, a knot formation was observed [Figure 3]. Complete removal of VPS and formal closure of the urinary bladder were performed, with the consequent placement of a delayed bladder probe, which remained in the patient for 10 days. Clinical evidence of peritonitis, meningitis,

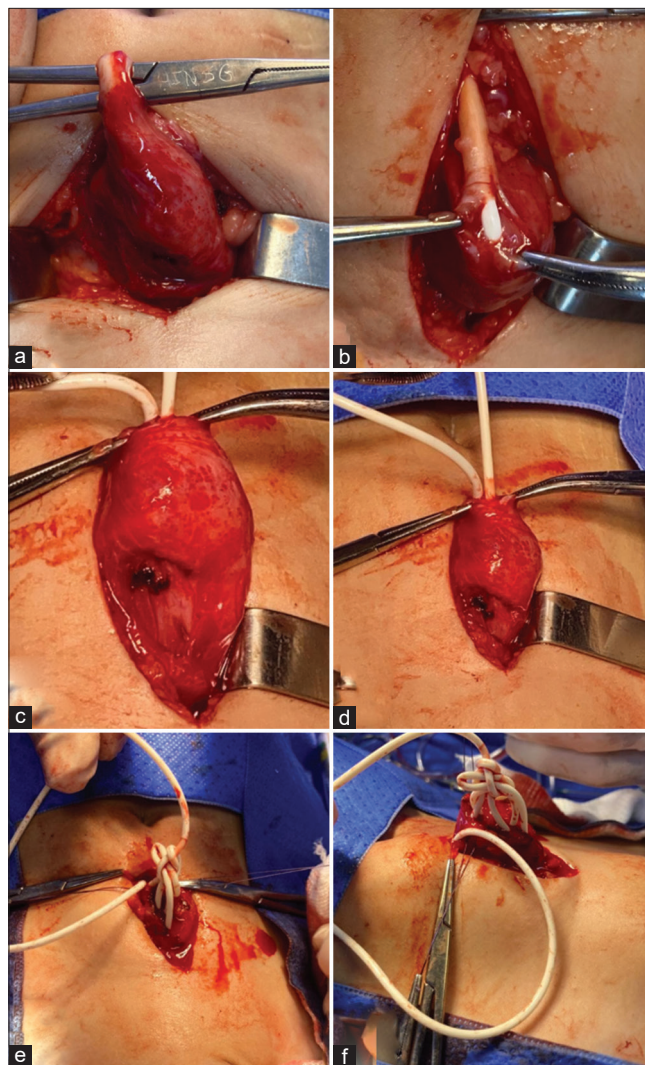


Figure 3: Intraoperative images showing the fibrous lining around the distal portion of the VPS that invaded the urinary bladder (a). Local dissection of the fibrous lining that circumscribed the VPS catheter, fixing part of it to the urinary bladder (b). Careful traction of the catheter to remove it from the inside of the bladder (c and d). After vesicostomy, the formation of an intravesical ball (e and f) was observed. VPS: Ventriculoperitoneal shunt.

or elevated intracranial pressure was not detected. No intravesical stone formation was observed.

Pohlman *et al.* (2011) and Kataria *et al.* (2013) reported two cases that describe two intravesical knot formations. However, in both cases, the clinical presentation was the extrusion of the distal portion of the VPS catheter through the urethra. This case presented only a clinical picture with chronic symptoms characterized by dysuria, urinary incontinence, intestinal constipation, and abdominal pain, symptoms that have been followed for a long time due to a history of myelomeningocele hindering a directed urgent approach. There were no signs and symptoms of meningitis or peritonitis.

The intraoperative findings showed a fibrous lining around the distal portion of the VPS that invaded the bladder [Figure 3]. It is believed, therefore, that the tip of the VPS catheter, initially and gradually, eroded the urinary bladder, puncturing it and provoking local irritation and inflammation, and inducing a localized reaction that culminated in the formation of a thin layer of fibers. Specifically, due to the long period of this process, the VPS catheter was surrounded by a fibrous coating at the perforation site, preventing the spread of urine and consequent peritonitis.

The possible causes for the formation of the rare and spontaneous intravesical knot that was found could be: effects of retained memory of the catheter, causing it to return to the position it was in the package when supplied; peristaltic bowel movements; increased intra-abdominal pressure; pulsation of cerebrospinal fluid; and pulsations of nearby blood vessels. However, the mechanisms by which the formation of intravesical knot occurs are still not fully understood.^[8]

CONCLUSION

Migration and perforation of the urinary bladder through the distal end of the VPS are quite unusual, unpredictable, and unavoidable conditions. The formation of intravesical knots is extremely rare and its mechanism of formation is still not well understood. Patients with VPS need adequate medical follow-up to identify possible complications of the shunt. Although it is a rare condition, perforation of the urinary bladder by the VPS catheter should be considered as a differential diagnosis in patients who, concurrently with the use of VPS, present urinary symptoms. It is recommended that urinary tract ultrasound and/or abdominal ultrasound are performed in these cases, guiding a more effective immediate management, thus avoiding a late diagnosis that leads to unwanted complications.

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Declaration of patient consent

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

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Nil.

Conflicts of interest

There are no conflicts of interest.

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