



CASE REPORT

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Ventriculo-peritoneal shunt trans-anal protrusion causing *Escherichia coli* ventriculitis in child: Case report and review of the literature

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Abstract

Background: Ventriculo-peritoneal shunting is an effective treatment for hydrocephalus, bowel perforation by peritoneal catheters is an extremely rare complications associated with this procedure.

Case presentation: We report a 9-year-old girl who had anal VP shunt protrusion two months following surgery and review the literature to understand the diagnosis and treatment for this rare complication. A PubMed search using the keywords 'bowel perforation' and 'Escherichia coli ventriculitis' was performed and citations were reviewed.

Conclusions: Neurosurgeons should be alert to early diagnosis and appropriate treatment of this condition to prevent infectious and neurological complications.

Background

Bowel perforation by peritoneal catheter (BPPC) is a kind of rare complication that could occasionally occur after ventriculo-peritoneal shunt (VPS) procedure and could result in potentially fatal ventriculitis or sepsis [1]. Patients suffering from BPPC do not always present with significant abdominal symptoms (less than 25%) such as peritonitis. Awareness and early recognition of this complication are essential because of the high mortality rate in such patients (15%) [2]. Until now, the literature across the world has been reported cases on 112 patients with BPPC, and more than half of the cases occurred within the age group of 0–10 years. Here, we report on a 9-year-old girl who had an anal VP shunt protrusion 2 months following surgery with *Escherichia coli* ventriculitis and with no signs of peritonitis, bowel obstruction, or VPS malfunction, and we review the literatures

on this rare but serious complication. Our goal is to alert physicians and neurosurgeons to the early diagnosis and treatment of this complication to minimize or prevent infectious and neurological consequences.

Case presentation

A 9-year-old girl diagnosed with germinoma in the third ventricle and obstructive hydrocephalus underwent VP shunting at the end of December 2015 in our hospital. She was given chemotherapy and noticed the catheter protruding through the anus one day before presentation in our emergency department in January 2016.

She presented with fever, headaches and vomiting. Physical examination showed she was alert but weak, and fundoscopy did not reveal papilledema. She exhibited signs of ventriculitis of neck stiffness, but she had no abdominal pain, her abdomen was soft, and no catheter was observed in the anal sphincter area or palpated during a rectal exam.

The abdominal X-ray did not show any shunt fracture or free abdominal air but confirmed the penetration of the distal (peritoneal) catheter of the VP shunt into the bowel as well as its trajectory from the abdomen towards the perineal region (Fig. 1). Computed

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Fig. 1 Left: Picture of the distal peritoneal catheter of the ventriculoperitoneal shunt protruding through the patient's anal region. Clear CSF drops could be seen at the end of the catheter. Right: Plain abdominal x-ray confirming bowel perforation by the distal peritoneal catheter as well as its trajectory from the abdominal cavity toward the perineal region

tomography (CT) of the abdomen revealed that the distal catheter penetrated the descending colon and followed the bowel towards the sigmoid colon (Fig. 2)

After the neurosurgery team discussion, the patient was operated to get the proximal (ventricular) catheter externalized. During the operation, an incision was created over the distal shunt on the abdominal wall. The peritoneal catheter was disconnected from the proximal portion and then removed from the distal portion without resistance. The proximal catheter was connected to a collecting bag. A post-operative abdominal ultrasound revealed no complications. Cerebrospinal fluid (CSF) analysis revealed a pattern suggestive of bacterial meningitis (leukocytes 5956/ul, glucose 1.7 mmol/L, and protein 117 g/L). The cultures of both the initial CSF sample and the cranial tip of the VP shunt revealed *Escherichia coli*, and treatment with the broad-spectrum antibiotic meropenem was initiated. CSF tests were performed daily. The child had a rapid, uneventful recovery without fever and CSF cultures became negative in ten days. After the end of the antibiotic treatment and three negative cultures, a repeated MRI scan revealed the tumour shrinkage and no signs of hydrocephalus after the distal catheter was clipped for over 48 h, and the whole VP shunt system was removed. The girl recovered uneventfully without additional antibiotic

treatments and get back to her baseline condition for the last 4 months.

She was in good condition, and a repeated MRI revealed no signs of the tumour after chemotherapy at the last follow-up.

Discussion

Bowel perforation caused by a VPS tube is pretty rare with an estimated incidence from 0.1% to 1.0% in shunted patients [3, 4]. However, this condition could cause devastating complications, such as meningitis and ventriculitis and carries a mortality rate of approximately 15% [2]. The first case of bowel perforation was reported in 1966 [5]. The shunt abdominal catheter may protrude from the large bowel [6], urinary bladder [7], urethra [8], or even the oral cavity [9]. The most common perforation site is the colon [10].

In a recently published review by Hai et al. [11], anal protrusions of distal shunt catheters were reported in 96 patients. Since that time, 15 cases have been reported [7, 8, 12–23]. In total, there are 112 cases including our case. Among these 112 cases, the ages of the 57 cases ranged from 0 to 10 years, and our patient belonged to this group. We think that the number of reported patients with this complication may have been higher if we had included articles published in non-indexed journals

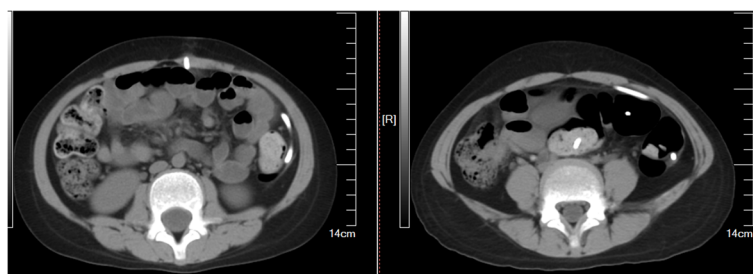


Fig. 2 Abdominal computed tomography scan demonstrating the migration peritoneal catheter in the bowel lumen

[13]. The interval between the shunt operation and the occurrence of the anal shunt protrusion ranges from weeks to several months. In our case, this interval was nearly two months.

More than 50% of BPPC patients are asymptomatic, and the most common complaint is protrusion of the shunt catheter through the anus [24]. Among those with a catheter through the anus, many patients were diagnosed in an asymptomatic stage (42%). The common symptoms are fever, abdominal pain, and vomiting (40%), and the less common presentations are shunt malfunction, an abdominal abscess, and peritonitis.

The exact pathogenesis is difficult to discern. Akyuz et al. [25] hypothesized that the shunt catheter tip adheres to the viscera wall, thus forming the constant pressure of the abutting tip along with a local inflammatory reaction, finally leads to the erosion of the visceral wall and tip enter into the lumen. The peristaltic activity of the gut carries the catheter all the way down to the anus. Because it is usually a localized the inflammation, so, there are rarely any obvious signs of peritonitis.

Many authors have reported the formation of an encasing fibrosis around the catheter both at surgery and at autopsy, which is thought to have an anchoring effect on the tube that results in pressure on an area of the bowel. Pressure on the bowel area may finally cause the bowel to erode and result in perforation.

There are some other mechanisms been suggested in the literature, including weakening of the intestinal wall in a poor general condition patient and the stiff end of the shunt tube causing the perforation.

The use of a trocar during surgery may predispose patients to the occurrence of this complication. Other techniques for inserting peritoneal catheters include mini-laparotomy and laparoscopic-assisted insertion [26].

Techniques to investigate BPPC include abdominal X-ray and CT [10, 27]. Plain x-rays may confirm spontaneous bowel perforation because they can demonstrate the trajectory of the peritoneal VP-shunt catheter towards the perineal region. Abdomen CT scans are helpful to rule out the presence of an abdominal abscess. X-rays, CT scans, and CSF cultures are positive in nearly 50% of cases.

In contrast, *E. coli* meningitis is also an indicator of intestinal perforation by a VP shunt tube [28].

Escherichia coli is the most common organism in BPPC cases. Based on the reported cases, the patients suffered from either ventriculitis or peritonitis, and some suffered both.

Once the diagnosis of BPPC has been established, the treatment should include total shunt removal, externalization of the ventricular drainage, and the broad-spectrum antibiotic therapy for at least 2 two

weeks or until CSF cultures from the ventricles are negative.

The shunt catheter should be disconnected at the abdominal wall, and the distant end should be removed from the rectum assisted by colonoscopy or sigmoidoscopy [15]. The distal end of the V-P shunt should not be pulled back into the peritoneal cavity to prevent contamination of the tract. Because the bowel perforation caused by the shunt is usually very small, in most of the cases, it would seals by itself. Thus, laparotomy is rarely necessary and is indicated only for those patients who present with acute abdomen [11, 29, 30].

The treatment for early and late perforation may be quite different. For very acute (within a day) cases, it probably need an emergent operation to suture the perforation through laparotomy, while in chronic cases (from weeks to months after surgery), it maybe enough to remove the shunt as our case. Overall, most cases recovered very well and do not need an abdominal surgery to treat the bowel perforation.

Fortunately, our patient was, such as the majority of reported cases, asymptomatic and without any abdominal symptoms or signs. We hypothesize that the distal catheter perforated the descending colon, passed through the distal bowel, and exited out of the anus.

An external ventricular drainage should be kept for at least for 3 weeks, and patient should be given broad-spectrum antibiotics to prevent/treat infections of the CSF. CSF infection due to the retrograde spread of the infection which may causing ventriculitis or meningitis should be ruled out after the extrusion of the shunt through the anus [19]. Once the CSF cultures are negative, a second shunting could be implanted if needed.

Antibiotics need to start to treat possible ventriculitis or meningitis causing by the retrograde spread of the infection prior to externalization [3, 31]. The antibiotics that have been reported include linezolid, meropenem and metronidazole. The use of a sequential combination of intravenous antibiotics and intraventricular antimicrobial therapy (IVT) therapy has also been reported [32].

The possible factors responsible for BPPC are thin bowel walls in children, a sharp and stiff end of the V-P shunt [29], the use of a trocar by the operating surgeons [33], chronic irritation from the shunt, previous surgery, infection and silicone allergy [34].

Because of their weak bowel musculature, children are more susceptible to intestinal perforation. The use of modern soft and supple catheters composed of silicone that may incite a lesser foreign body reaction has been said to decrease the incidence of this complication.

Other techniques for inserting the peritoneal catheter, such as mini-laparotomy and laparoscopic-assisted insertion, may be helpful to prevent BPPC. However, large retrospective series have failed to demonstrate an advantage of one technique over the other in routine use [26].

Conclusion

We described the case of a 9-year-old girl with an anal protrusion of a VP shunt that presented 2 months following the shunt placement. Based on our case and a literature review, we stress the following issues: (a) the importance of early diagnosis and treatment to minimize or prevent infectious and neurological complications [7, 35], (b) the externalization of the proximal edge of the shunt without pulling the distal tip proximally by cutting the distal shunt tube over the abdominal wall and pulling the distal edge from the anal side to prevent the spread of infection spread, (c) the application of broad spectrum antibiotics until the CSF cultures are negative, and (d) the focus of attention on the abdominal symptoms and signs.

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Availability of data and material

Not applicable.

Authors' contributions

CL and YT conceived and designed the study. YL and YT wrote the paper. All authors read and approved the manuscript.

Competing interests

The authors declare that they have no competing interests.

Consent for publication

Informed consent was obtained for submission and publication of the case report from the parents of the respective patients.

Ethics approval and consent to participate

For this type of study formal consent is not required.

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