CASE REPORT

Ileal perforation and transanal protrusion of the peritoneal tube in a boy with a ventriculoperitoneal shunt and literature review

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Abstract
Ventriculoperitoneal shunting (VPS), a widely used procedure for treating hydrocephalus, may cause various complications, including mechanical failure, shunt infection, and intra-abdominal complications. Among these, intestinal perforation is rare. Patients suffering from intestinal perforation may be asymptomatic or present symptoms, such as abdominal pain, vomiting, fever, abdominal abscess, and peritonitis. However, such patients rarely manifest transanal protrusion of the peritoneal tube, which results in bowel perforation in the colon. In this report, we present the case of a 3-year-old boy with VPS-induced small-intestinal perforation and peritoneal-tube transanal protrusion. Additionally, a review of the literature on VPS-induced small-intestinal perforations revealed no similar cases.

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1. Introduction

Ventriculoperitoneal shunting (VPS) is the most widely used procedure for treating hydrocephalus. 1 Although it is effective and safe, VPS may cause various complications, such as shunt obstruction, catheter disconnection or loss, intestinal obstruction, inguinal hernia, ascites, intestinal volvulus, bowel perforation, extrusion through the umbilicus or abdominal incision, and pseudocyst formation, and has a complication rate of 24–47%. 1–4 Intra-abdominal
complications account for ~10–30% of all complications.\(^2,3\) Intestinal perforation is a rare VPS complication, with an incidence estimated at 0.1–2.5%.\(^1,4\) Patients experiencing intestinal perforation may be asymptomatic or present symptoms, such as abdominal pain, vomiting, fever, shunt malfunction, abdominal abscess, and peritonitis. Furthermore, the distal end of the peritoneal tube may migrate to the heart, urethra, or anus through the bowel or umbilicus.\(^1\) Among these migrations, transanal protrusion of the peritoneal tube is rare, but commonly recognized, and facilitates the diagnosis of intestinal perforation.\(^1,4\) Additionally, all patients manifesting transanal protrusion of the peritoneal tube have a bowel perforation in the colon.\(^2,3\) In this report, we present the case of a 3-year-old boy with a small-intestinal perforation and peritoneal-tube transanal protrusion 18 months after VPS. A literature review revealed that this is the first report of such a case.

2. Case report

A 3-year-old boy with a history of traumatic subarachnoid hemorrhage and skull fracture at age 1.5 months developed hydrocephalus and underwent VPS at 9 months. At 18-months old, the VP shunt was replaced with a new shunt because of infection. Eighteen months later, he experienced abdominal pain for 3 days, and his mother discovered a tube protruding from his anus following defecation (Figure 1). The abdominal pain was intermittent and typically resolved spontaneously after several hours. He did not experience fever, chillness, nausea, vomiting, or melena. On admission, he had clear consciousness, stable vital signs, a body temperature of 36.8°C, and no neck stiffness. He had mild epigastric tenderness, but did not display any peritoneal signs. A 10-cm yellowish tube was protruding from the anus, with clear fluid draining from it. A complete blood count showed hemoglobin of 12.0 g/dL and a white blood-cell count of 7.12 × 10^3/μL, with 41.2% neutrophils and 49.6% lymphocytes. Serum biochemistry and cerebrospinal fluid (CSF) examinations were normal, and the CSF culture yielded no growth. A plain abdominal roentgenogram revealed focal ileus at the mid-abdomen and the peritoneal tube of the VP shunt encircling the abdomen and progressing to the perineal region (Figure 2A). Abdominal computed tomography (CT) revealed that the tube penetrated one segment of the small intestine and progressed distally in the colon (Figures 2B and 2D). Free air or ascites were not observed. We performed laparoscopic exploration by administering broad-spectrum antibiotics, because of the focal ileus and because the patient continued to experience intermittent abdominal pain. Initially, the proximal part of the peritoneal tube of the VP shunt was cut at the right side of the neck. Subsequently, laparoscopy was performed, during which tight adhesion was observed in the abdomen. A thick fibrous bundle encasing the peritoneal tube extended from the right upper abdominal wall to the terminal ileum, with adhesion between the peritoneal tube and the ileum. The fibrous bundle entered the terminal ileum following lysis of the adhesion 25 cm proximal to the ileocecal valve. The fibrous bundle and the terminal ileum were subsequently withdrawn from the laparoscopic port wound at the umbilicus. After removal of the fibrous tissues, the peritoneal tube was excised at its ileum entry, the distal end was withdrawn from the anus, and the proximal end was removed from the ileum extracorporeally. The fibrous tissue and perforation site were excised, and the ileum was repaired extracorporeally. Antibiotic treatment was continued after surgery, and the patient recovered well. He was discharged 6 days after surgery, and the oral antibiotic treatment was continued for 2 weeks. Later, his shunt was further managed by a neurosurgeon half a year later.

3. Discussion

Among VPS-induced gastrointestinal perforations, colonic perforation constitutes the majority, whereas perforations of the stomach and small intestine are rare, with small-intestinal perforation being the rarest.\(^1,3\) Additionally, all patients manifesting peritoneal-tube transanal protrusion have been diagnosed with colonic perforation.\(^1,9\) Our case is the only report of small-intestinal perforation and peritoneal-tube transanal protrusion. Such bowel perforations are associated with fibrosis encasing the tube, which anchors the tube and exerts pressure on the bowel area, due to foreign-body reaction.\(^1\) This pressure coupled with the continuous hammer effect of CSF pulsations may eventually cause the bowel to erode, resulting in the perforation.\(^1\) Moreover, a thick fibrous bundle encasing the peritoneal tube and extending from the right upper abdominal wall to the terminal ileum was observed in our patient.

Only seven cases of VPS-induced small-intestinal perforation have been reported (Table 1).\(^3,5–9\) The perforation sites were the jejunum in three patients (proximal jejunum, middle jejunum, and an unknown location in the jejunum), ileum in two patients (25 cm and 75 cm proximal to the ileocecal valve), and unknown locations in the small intestine in two patients. The mean interval between shunting and the onset of symptoms was 16.3 months (range, 2.5 months–3 years). Furthermore, the clinical
manifestations were as follows: shunt dysfunction and CNS infection in two patients, and peritonitis, intestinal obstruction, and skin erosion with shunt-tube infection and transoral protrusion in one patient each. The remaining two patients died of congestive heart failure and recurrent medulloblastoma with CNS infection; their intestinal perforations were diagnosed at autopsy. The intestinal perforations in the surviving patients were diagnosed by injecting contrast medium into the shunt tube (shuntogram) in one patient, surgery in two patients, and shuntogram and surgery in two patients. The five surviving patients underwent surgery, which predominantly consisted of peritoneal-tube removal and proximal diversion, followed by VP-shunt revision. Additionally, two patients underwent laparotomy, with one patient each underwent intestinal resection and volvulus derotation with intestinal perforation closure. Our patient was a 3-year-old boy who had posttraumatic hydrocephalus, and presented with peritoneal-tube transanal protrusion and abdominal pain. The perforation site, diagnosed at surgery, was in the terminal ileum, and the interval between the latest VPS and the subsequent perforation was 18 months. He underwent laparoscopic exploration for extracting the distal and proximal ends of the peritoneal tube from the anus and extracorporeally from the ileum, respectively, after which the ileal perforation was closed. The clinical characteristics and management of the seven patients with small-intestinal perforation and our patient were similar to

Figure 2  (A) Plain abdominal roentgenogram revealed focal ileus at the mid-abdomen, and the peritoneal tube encircled in the abdomen and progressing to the perineal region. (B) Abdominal computed tomography revealed a radiopaque tube entering the small intestine. (C) The tube is in the ascending and descending colon. (D) The tube progresses along the whole transverse colon.
<table>
<thead>
<tr>
<th>Case</th>
<th>Age/sex</th>
<th>Cause of initial shunt operation</th>
<th>Interval between onset of symptoms &amp; last VPS</th>
<th>Clinical manifestations</th>
<th>Perforation sites/diagnostic method</th>
<th>Treatment</th>
<th>Outcome</th>
<th>Reference</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>4 y, 4 mo/F</td>
<td>Aqueductal stenosis</td>
<td>2.5 mo</td>
<td>CHF (+)</td>
<td>Proximal jejunum/autopsy</td>
<td>No</td>
<td>Death</td>
<td>[3]</td>
</tr>
<tr>
<td>2</td>
<td>3 y, 2 mo/F</td>
<td>Medulloblastoma with hydrocephalus</td>
<td>3 mo</td>
<td>Recurrent medulloblastoma, CNS infection</td>
<td>Ileum (75 cm from ICV/autopsy</td>
<td>No</td>
<td>Death</td>
<td>[5]</td>
</tr>
<tr>
<td>3</td>
<td>3 y/F</td>
<td>Communicating hydrocephalus</td>
<td>3 y</td>
<td>VP shunt dysfunction</td>
<td>NM/shuntogram and surgery</td>
<td>Removal of peritoneal tube and insertion of new tube</td>
<td>NM</td>
<td>[6]</td>
</tr>
<tr>
<td>4</td>
<td>29 y/M</td>
<td>Congenital hydrocephalus</td>
<td>2 y</td>
<td>Meningitis, VP shunt malfunction</td>
<td>NM/shuntogram</td>
<td>Removal of VP shunt and insertion of a new one</td>
<td>NM</td>
<td>[7]</td>
</tr>
<tr>
<td>5</td>
<td>5 y, 9 mo/M</td>
<td>Posttraumatic hydrocephalus</td>
<td>2 y, 8 mo</td>
<td>Peritonitis, meningitis</td>
<td>Jejunum/shuntogram &amp; surgery</td>
<td>Laparotomy (resection of jejunum with proximal diversion), VPS revision</td>
<td>NM</td>
<td>[8]</td>
</tr>
<tr>
<td>6</td>
<td>11 mo/M</td>
<td>Hydrocephalus, meningomyelocele</td>
<td>11 mo</td>
<td>Fever, vomiting, abdominal distension, volvulus</td>
<td>Ileum (25 cm from ICV (with volvulus)/laparotomy</td>
<td>Laparotomy (derotation of volvulus, closure of ileal perforation, shunt removal, EVD), VPS revision</td>
<td>Good</td>
<td>[9]</td>
</tr>
<tr>
<td>7</td>
<td>1 y, 3 mo/F</td>
<td>Hydrocephalus</td>
<td>6 mo</td>
<td>Skin erosion, shunt infection, transoral protrusion of tube</td>
<td>Middle jejunum/laparotomy</td>
<td>Removal of peritoneal tube</td>
<td>Good</td>
<td>[9]</td>
</tr>
<tr>
<td>This case</td>
<td>3 y/M</td>
<td>Posttraumatic hydrocephalus</td>
<td>1 y, 6 mo</td>
<td>Abdominal pain, transanal protrusion of tube</td>
<td>Ileum (25 cm from ICV/laparoscopy</td>
<td>Removal of peritoneal tube, laparoscopic closure of ileal perforation</td>
<td>Good</td>
<td></td>
</tr>
</tbody>
</table>

CHF = congestive heart failure; EVD = external ventricular drain; ICV = ileocecal valve; NM = not mentioned; Shuntogram = injection of contrast medium into the VPS; VPS = ventriculoperitoneal shunt.
those of the patients with VPS-induced bowel perforation in other locations.

VPS-induced small-intestinal perforation is rarer than colonic perforation. This is likely because the colon is fixed, whereas the small intestine is mobile in the abdomen, which renders peritoneal tube-induced fibrosis difficult to fix to the small intestine and results in bowel perforation similar to colonic perforation. Furthermore, the duodenojejunal junction and the ileocecal junction are relatively fixed compared with other sections of the small intestine. Thus, VPS-induced bowel perforation may more commonly occur in these sections of the small intestine, similar to our case. Additionally, considering the large separation between the terminal ileum and the anus, transanal protrusion of the peritoneal tube is difficult. However, the plain abdominal roentgenogram of our patient revealed that a long section of the peritoneal tube was present in the abdomen, indicating that an extended peritoneal tube was used during VPS for eliminating the need to lengthen the peritoneal catheter as the patient aged. The length of the peritoneal tube was crucial in the transanal protrusion observed in our patient.

The diagnosis of VPS-induced bowel perforation is often difficult and predominantly depends upon the clinical manifestations, abdominal X-ray, ultrasound, and CT results. Typically, bowel perforation is considered in shunted patients with unexplained fever or prominent abdominal symptoms. Additionally, gut flora meningitis, confirmed through CSF culture or pneumocephalus observed in the head CT, may be an indicator of intestinal perforation. However, many cases do not manifest shunt infection or peritonitis, and bowel perforation is recognized only during shunt revision for obstruction. By contrast, the diagnosis is easy if patients with prior VPS operation present with anal protrusion of the tube, as was the case in our patient. The plain abdominal roentgenogram revealed that the long peritoneal tube encircled the abdomen and progressed to the perineal region, and the CT scan revealed that the tube entered the ileum and progressed to the colon. Occasionally, a shuntogram can be used to demonstrate the mucosal pattern of the intestine, which was the case in the three patients with VPS-induced small-intestinal perforation reported in the literature.

The management of the VPS-induced small-intestinal perforation must be individualized according to the clinical conditions of the patients. Typically, removing the protruded shunt system and controlling the infection, followed by CSF diversion, is the standard treatment protocol, however, abdominal exploration for managing VPS-induced intestinal perforation is controversial. Several reports demonstrated that the peritoneal tube can be removed without abdominal exploration, because the opening into the bowel lumen is often small and self-sealing in the absence of peritonitis or abdominal abscess. Abdominal exploration for perforation repair might be required in cases accompanied by peritonitis, abdominal abscesses, acute bowel perforation, failure of the closure of the fistulous tract following peritoneal tube removal, difficulty in removing the peritoneal tube, and knotting or twisting of the peritoneal tube. In a report reviewing 45 cases of VPS-induced intestinal perforation, the shunt tube was removed without abdominal exploration in 31 patients (68.9%), laparotomy for bowel repair was performed in eight patients (17.8%), and an unclear method of tube retrieval was used in six patients (13.3%). Concerning abdominal exploration, laparoscopic surgery can be performed as an alternative to laparotomy, however, only in one report was a patient with VPS-induced silent bowel perforation managed laparoscopically. Our patient had persistent intermittent abdominal pain and focal ileus; therefore, we performed laparoscopic surgery for diagnosis and definite repair. The resection of the involved ileum and Anastomosis was performed extracorporeally through laparoscopy.

The prognosis for recovery in patients with VPS-induced bowel perforation is more accurate when bowel perforation is detected at the asymptomatic stage. However, if bowel perforation is undetected, patients are at a high risk of developing meningitis or ventriculitis, and the mortality rate may increase to 15%.

In conclusion, ileal perforation and transanal protrusion of the peritoneal tube following VPS is rare. Management should be individualized according to the clinical situation. Most patients may be treated through shunt removal, external ventricular drainage, and antibiotic treatment, whereas some may require abdominal exploration. For abdominal exploration, laparoscopic surgery is an alternative to laparotomy, because it provides clear diagnosis. Moreover, through laparoscopy, the peritoneal tube can be removed and definite repair can be performed extracorporeally without the risk of spreading the infection.

References