Case Report

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Bowel perforation after ventriculoperitoneal-shunt placement: case report and review of the literature

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Abstract: Bowel perforation by a peritoneal catheter (BPPC) is a serious complication after ventriculoperitoneal shunting, with high mortality and morbidity rates. This patient presented with scalp ulceration over the shunt valve at the retromastoid region 26 years after shunt placement. During revision, the catheter distal to the valve was divided in the clavicular region. As there was no cerebrospinal fluid drainage, we decided to remove the ventricular catheter and valve. The ulceration was debrided and primarily closed. Distal to the clavicle, the disconnected peritoneal catheter was encased in a fibrous, calcified tunnel. To avoid piecemeal resection with multiple incisions, the catheter was not retrieved. Two years later, the patient presented with an abscess and pus draining from the clavicular wound. Cultures were positive for enteric bacteria. BPPC with retrograde spread of infection was suspected, and abdominal computed tomography confirmed perforation. We removed the disconnected catheter, but the perforation site could not be detected during laparotomy. The patient was treated with intravenous antibiotics and recovered without complications. To minimize complications, abandoned catheters should be avoided. Otherwise, patients unnecessarily have a life-long risk of developing abdominal complications. In patients with abandoned catheters and severe abdominal symptoms, BPPC is an important differential diagnosis.

1 Introduction

Hydrocephalus remains a serious condition that is characterized by pathological production or absorption of cerebrospinal fluid (CSF) in the brain [1]. The clinical presentation may be various and depends on the degree of brain damage caused by increased intracranial pressure [2]. To avoid intracranial pressure induced deficits or death and to improve the clinical outcome of these patients, ventriculoperitoneal (VP) shunting remains the surgical standard in the treatment of congenital hydrocephalus [2-4].

Bowel perforation by a peritoneal catheter (BPPC) [5] is a rare but serious complication after VP shunting [5, 6]. In a large series, the incidence of BPPC was 1% and mortality rates of up to 15% have been reported [5, 6]. Although the diagnosis is evident in cases with anal catheter protrusion, perforations may unexpectedly be recognized during shunt revision for obstruction or infection [5]. Interestingly, <30% of patients with BPPC present with signs of peritonitis [6].

During revision, total shunt removal may be difficult in some cases. Metal connectors placed after piecemeal shunt revision can lead to increased difficulty with removal [7]. Furthermore, it has been demonstrated that shunt catheters become degraded during long-term implantation leading to calcifications or are encased in a fibrous reaction zone called a fibrous tunnel [8]. To avoid piecemeal resection, the peritoneal and non-draining catheter is frequently left within the abdomen, if it cannot be easily removed. In the literature, such distal segments are referred to as abandoned, disconnected, orphaned, or redundant shunt catheters [9, 10].

We have recently reported the causes of peritoneal catheter revisions in patients treated for pediatric hydrocephalus with follow-up of at least 20 years. Only one out

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of 112 (0.9%) patients was treated for BPPC [11]. In that case, during operation for suspected shunt infection, occult bowel perforation was unexpectedly detected. This patient was successfully treated with initial shunt externalization and then placement of a new peritoneal catheter [11].

Only a very few cases of intraabdominal complications from abandoned distal catheters have been reported so far [7]. However, due to the reported significant morbidity and mortality rates after BPPC [5, 6], strategies to avoid abdominal complications especially in long-term survivors are highly desired. Here, we present additionally a rare case of BPPC in a patient included in the above-mentioned study cohort with an abandoned peritoneal catheter that occurred 28 years after the initial VP shunt placement.

2 Case report

In this female patient with a meningomyelocele (MMC) a ventriculoatrial (VA) shunt was inserted at the age of 15 months (February 1984). A short atrial catheter was replaced by a peritoneal catheter after six years.

Twenty-six years (June 2016) later, this patient presented with scalp ulceration above the shunt valve at the retromastoid region. The shunt valve and distal catheter were visible. There were no other signs of infection (meningism or fever). Inflammatory parameters, such as C-reactive protein were within the normal range.

During shunt revision, the catheter distal to the valve was first divided in the clavicular region. Given that there was no cerebrospinal fluid drainage, this patient was regarded to be shunt-independent. Therefore, we decided to remove the ventricular catheter and shunt valve. The ulceration was debrided and primarily closed. Distally to the clavicle, the disconnected peritoneal catheter was encased in a fibrous, calcified tunnel. To avoid piecemeal resection with multiple skin incisions, the catheter was left in situ and was not retrieved.

Two years later (July 2018), the patient presented with a subclavicular abscess and pus draining from the clavicular wound. There were no other signs of infection (fever, meningism, diarrhea, peritonitis). Bacterial examinations revealed cultures positive for *Escherichia coli* and *Enterobacter*.

Bowel perforation by the distal peritoneal catheter with retrograde spread of infection was therefore suspected, and abdominal computed tomography (CT) confirmed perforation with presence of the distal catheter in

the colon descenders (Figure 1). There were no radiological signs of free air or abscess in the abdominal cavity.

The disconnected distal catheter was completely removed, and exploratory mini-laparotomy was performed by an experienced general surgeon. The perforation site could not be detected, but the distal catheter was found to be fecally stained (Figure 2). Detailed bacterial staining of the distal catheter re-confirmed the growth of *E. coli* and *Enterobacter*. The subclavicular wound was primarily closed. This patient was additionally treated with intravenous antibiotics and completely recovered with no subsequent postoperative complications.

Ethical approval: Research has been complied with all the relevant national regulations, institutional policies and in accordance the tenets of the Helsinki Declaration.

Informed consent: Informed consent has been obtained for all surgical procedures and investigations.

3 Discussion

In this paper, we present a rare case of BPPC in a patient with an abandoned peritoneal catheter 28 years after initial VP shunt placement. In light of the overall high mortality and morbidity rates [5-7], BPPC can be regarded as a significant long-term complication. The overall incidence of BPPC in the long-term follow-up of our patient cohort is 1.8% (2 out of 112 patients). A similar incidence (1%) was found in a large study, where 1956 patients were followed for a mean duration of 10 years [5]. Given that BPPC even occurs in long-term survivors, its incidence may be dependent on the length of follow-up.

Beside BPPC, non-enteric visceral perforations, including the urinary bladder, vagina, gall bladder, stomach, scrotum, liver, or uterus, have been reported [5, 12].

The etiology of BPPC remains unknown in many cases. However, several risk factors have been postulated including the thin bowel wall of children, stiff distal catheters, silicone allergy, or use of abdominal trocars [13]. The mechanisms of BPPC may be categorized into the following two types: an acute-traumatic type occurring during the initial distal catheter placement or a chronic-irritative type [7]. The chronic-irritative type may be the main reason for late cases of BPPC and is caused by a break in the continuity of the epithelium around the contact site of the distal catheter leading to local inflammation, fibrosis, and adherence of the catheter to the bowel wall. Continuous cerebrospinal fluid pulsations in combination with peristalsis and patient motion may then contribute to per-

Figure 1: Native abdominal computed tomography scan indicates bowel perforation. Axial (A) and coronal (B) views. White arrow indicates the distal end of the ventriculoperitoneal shunt catheter inside the colon descends.

Figure 2: Fecally stained peritoneal catheter. * indicates the distal end (A); the peritoneal catheter was divided after removal (B); the arrow indicates intraluminal feces.

foration of the bowel wall [7, 10, 12]. A fibrous encasement of the distal catheter at the enterotomy side may occur without a subsequent peritoneal infection. Therefore, the diagnosis of BPPC can be difficult, delayed, and sometimes unexpectedly recognized during shunt revision [5]. Although the exact etiology remains unknown, the chronic-irritative type is the most likely mechanism of BPPC in our patient.

Sathyanarayana reviewed 45 cases of BPPC and found out that 42% of patients were asymptomatic at diagnosis. Presentation of BPPC included anal extrusion of catheter (44%), abdominal symptoms (29%), fever (27%), shunt dysfunction (16%), or meningitis (13%) [6].

BPPC as a long-term complication (time from placement to BPPC diagnosis > 2 years) occurred in 7 (15.6%) out of these 45 reviewed cases. Three patients presented with anal protrusion of the catheter and two patients had abdominal pain and fever. One patient presented with

scalp ulceration over the valve and another patient with shunt malfunction. In no patient, BPPC occurred >8 years after shunt placement [6]. However, symptomatic BPPC > 20 years after initial shunt placement is extremely rarely reported [14].

Recently, in a large series, it could be demonstrated that 19 out of 1956 shunt treated patients were affected from BPPC. In this cohort, the median interval between last shunt revision and the diagnosis of BPPC was 2.4 years (range 5 days – 10.3 years). Interestingly, in 8 patients (42% of BPPC cases), BPPC was diagnosed more than 12 months after last revision. However, 6 out of these 8 patients presented with meningitis. These results clearly indicate, that BPPC occurring >12 months after last revision, frequently presents with signs of retrograde spread of infection [5].

Several other studies have described a retrograde spread of enteric organisms due to bowel perforation

leading to intracerebral sepsis including ventriculitis or meningitis [6, 15]. Alves *et al.* reported a case of symptomatic colonic perforation 28 years after the initial shunt placement presenting with acute meningitis. In this case, during endoscopic exploration, chronic fibrous tissue around the site where the catheter had entered the colon was observed; therefore, the spillage of bowel contents into the peritoneal cavity was prevented [14]. These papers indicate that patients with intracerebral sepsis due to enteric bacteria must be assessed for BPPC [6, 15].

Thiongo *et al.* report on a case where the retrograde spread of infection led to an enterocutaneous fistula, wherein the subcutaneous abscess moved upward from the intestine to the chest [13]. Similarly, in our case, the subclavicular abscess was caused by the retrograde spread of infection. This patient never presented with signs of meningitis or ventriculitis. Additionally, the abscess occurred 2 years after the distal catheter was disconnected. Therefore, it may be suspected that BPPC happened after removal of the ventricular catheter and shunt valve. It remains unknown, if BPPC would have been prevented with immediate removal of the peritoneal catheter.

Abdominal complications from abandoned catheters have been rarely reported, and a case with subcutaneous emphysema due to air moving from the perforated bowel through the intraluminal catheter has been described. In this patient, multiple jejunal perforations were observed during revision leading to partial small-bowel resection and re-anastomosis [7].

This study indicates that morbidity from abandoned catheters may be significant. Although patients with abandoned catheters are not at risk for developing meningitis, late diagnosis of peritonitis from BPPC may carry a mortality rate [7]. Therefore, complications from abandoned catheters should not be underestimated.

Principally, early as well as late BPPC must be regarded as an emergency [5], and surgical strategies depend on the clinical situation. In patients without peritonitis, abdominal exploration and localization of the entry site of the peritoneal catheter may not be necessary [7]. A laparotomy is primarily recommended for patients with an acute abdomen [5]. If it is not possible to remove the distal catheter, the perforated segment of the bowel may require resection and re-anastomosis [7]. According to a review involving 45 cases, the majority of peritoneal catheters could be directly extracted percutaneously, whereas, in 17% of cases, laparotomy and surgical repair of the bowel were required [6]. In many cases, the perforation site may seal off spontaneously, especially in cases without peritonitis. In addition, ventriculitis or meningitis is a life threatening neurosurgical emergency that must be

treated with complete shunt removal in combination with placement of an external ventricular drainage, if necessary.

Our patient presented without abdominal signs. The distal catheter was easily removed and fecally stained. During mini-laparotomy performed by an experienced general surgeon, the perforation site could not be detected and the patient recovered without further complications.

Further, our patient remained shunt independent during the follow-up. Recently, it has been demonstrated that 88% of patients with MMC require placement of a CSF draining shunt [16]. Although shunt surgeries are most common during the first years of life in patients with a MMC, it has been demonstrated that revisions are still necessary as late as 43 years of age [17]. Similarly, we could recently demonstrate that 2.8% of shunt revisions are performed twenty years after first shunt placement. In our long-term survivors, 14.3 % of patients are regarded as independent of a prosthetic shunt system [3]. These results are in line with another study that demonstrated a 12% shunt independency rate in long-term survivors after forty years [18].

In conclusion, to minimize complications, we recommend that abandoned catheters should be avoided whenever possible or at least the intraperitoneal part should be removed. Otherwise, these patients unnecessarily have a life-long risk for developing abdominal complications. In patients with abandoned catheters and severe abdominal symptoms, BPPC is an important differential diagnosis.

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