Bowel Perforation and extrusion of a ventriculo-peritoneal shunt through the anus

Perfuração Intestinal e Extrusão de Catéter de Derivação Ventrículo-Peritoneal Via Anal

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RESUMO

Derivação Ventrículo-Peritoneal (DVP) é um dos procedimentos mais frequentemente realizados para o tratamento de hidrocefalia. Entretanto, mesmo sendo tecnicamente simples e bem conhecido, existem várias complicações graves que podem ocorrer, sendo a perfuração intestinal uma delas. Esta complicação é rara, especialmente entre adultos e geralmente ocorre dentro do primeiro ano após o procedimento cirúrgico. Esta pode ainda, ser agravada, tanto por infecção, do sistema nervoso central ou sistêmica, quanto por aumento da pressão intracraniana devido ao mau funcionamento do sistema de drenagem. O quadro clínico geralmente oligossintomático muitas vezes torna o diagnóstico difícil, sendo necessário o uso de múltiplos exames complementares. Existem também inúmeras dúvidas quanto à fisiopatologia e fatores predisponentes para esta complicação. Ainda, devido à sua baixa incidência, sendo descrita, em sua maioria, através de relatos de caso ou pequenas séries, não existe consenso para seu ideal manejo. O tratamento varia desde abordagens menos invasivas, preservando os componentes da DVP, até a retirada completa de todos os componentes e uso de antibióticos de amplo espectro. Relatamos um caso de perfuração intestinal e extrusão de cateter de DVP através do ânus em um paciente adulto, tardiamente, discutindo os dados da literatura sobre este assunto.

Palavras Chave: derivação ventriculo-peritoneal, perfuração intestinal, extrusão anal, catéter

ABSTRACT

Ventriculo-peritoneal shunt (VPS) is one of the most commonly used procedures in the treatment of hydrocephalus. Nevertheless, even being technically simple and well-known, there are several serious complications that can happen, and among them is bowel perforation. This complication is rare, especially in adults, and it usually happens within the first year after the surgical procedure. It can also be aggravated by both infections, in the central nervous system or systemic, and also by increase in the intracranial pressure, due to shunt system dysfunction. The symptoms are usually mild, what can make the diagnosis challenging, and demanding several complementary tests. Also, there are many questions about the pathophysiology and predisposing factors for this complication. Due to its low incidence, and because it is usually described through case reports and small series, there is no consensus regarding its ideal management. The treatment varies from less invasive approaches, preserving the shunt components in place, until the full withdrawn of the whole system and use of wide spectrum antibiotics. We report a delayed case of bowel perforation and catheter extrusion through the anus in an adult patient, discussing the data available about this pathology.

Keywords: Ventriculo-peritoneal shunt complication, bowel perforation, anal extrusion of catheter

INTRODUCTION

Ventriculo-peritoneal shunts (VPS) are amongst the most frequently performed operations in the management of hydrocephalus. The incidence of complications is reported to be 5–47%. Bowel perforation is an unusual but serious complication of VPS and can be manifested by extrusion of VP shunt catheter from anus. Until 2000, less than 50 cases of bowel perforation with extrusion of catheter had been reported in the literature, to which few more cases have been added as case reports. The purpose of the present study is to show a case of an adult patient with bowel perforation and extrusion of the VPS through the anus and discuss about the available data about this issue.
We present the case of a male patient, 26 years of age, who had been submitted, 7 years earlier, to ventriculo-peritoneal shunting due to hydrocephalus secondary to an arachnoid cyst in the posterior fossa. The patient was admitted in the emergency room complaining about unspecific headache and reported fever. The neurological examination revealed only nuchal rigidity. There was no abdominal complaints or other abnormalities in the physical examination. A Computed Tomography (CT) scan of the brain was then performed and evidenced the arachnoid cyst in the posterior fossa, but excluded both hydrocephalus or signs of infection. In the seventh day of hospitalization, while waiting for some results of routine exams, the patient extruded the catheter through the anus, leaving exposed around 8 cm of its distal part (Fig. 1 a). There was a clear fluid (Cerebro Spinal Fluid, CSF) coming out actively from the tip of the catheter. The fluid was collected and taken to analysis. It showed no signs of infection and the culture was negative. The patient was then submitted to an abdominal CT scan which showed a perforation of the descendent colon, and the catheter inside of it. No abscess or other signs of infection were noticed (Fig 1 b, c). The patient was then submitted to laparotomy, and it was noticed involvement of the catheter by the omentum in about 5 cm, and fistulization to the descendent colon (Fig. 2). There was no bowel content inside the abdominal cavity. It was then performed a meticulous dissection of the omentum, section in the catheter above the level of entrance in the colon, primary suture of the bowel and repositioning of the remainder of the catheter. After 5 days, the intra-luminal distal catheter was eliminated spontaneously. The patient had a good recovery, but the CSF culture collected during surgery showed mixed bacterial (Escherichia coli) and fungal infections. Although the patient was still without any symptoms, we decided to withdraw the whole VP-Shunt system, to implant an external ventricular drainage (EVD) system and start antibiotics. The EVD system was withdrawn after 5 days. The patient remained in the hospital for 35 days, received standard antibiotics for post-operative CNS infection (Meropenen plus Vancomycin) and additionally Fluconazol. He was discharged without any symptoms and remained without the need of shunting until the present day.

**DISCUSSION**

The use of peritoneal cavity for CSF absorption in VPS was introduced in 1905, by Kausch. Since then VPS is amongst the most frequently performed operations in the treatment of hydrocephalus. Other shunting techniques are ventriculo-atrial shunt, lumbo-peritoneal shunt, third ventriculostomy. Although the risk in performing a shunt operation is low, the complications related to shunts are many, with a reported incidence of 24–47%. Malfunction of the VPS occurs in 25–35% of patients at 1 year, and 70–80% of patients require at least one revision at some point in their lives. The most common causes of shunt malfunction are catheter obstruction
and infection⁴. In these 2 groups, it can be cited infection of the shunt, malfunction due to blockage, disconnection, migration, equipment failure, extra-peritoneal retraction of the catheter, incisional hernia, subcutaneous collection of CSF and peritoneal pseudocyst formation, the latter due to low-grade infection followed by wrapping by the omentum. Other complications reported include intestinal perforation, CSF ascites, inguinal hernia, and intestinal volvulus⁴.

These complications may manifest as local abdominal signs, increased intracranial pressure or as an infectious processes⁴. Shunt infection remains a frequent and potentially fatal complication of CSF diversion with reported incidence of 5–10% and approximately 70% of shunt infections manifest within 2 months after shunt insertion⁷. Abdominal complications are reported to occur in 25% of VPS¹. Intra-abdominal complications after VPS placement are most commonly located near the peritoneal end of the shunt catheter; more than 50% of patients require shunt revision⁴. Visceral perforation is an unusual but serious complication of VPS with a reported mortality of up to 15%⁷. Wilson et al. first reported an intestinal perforation induced by distal shunt catheter in 1966¹⁷. The bowel is the most commonly involved site for perforation and is reported to occur in less than 0.1–0.7% of cases, and nearly half are diagnosed after removal of the catheter⁴⁷. This complication can lead to fatal central nervous system (CNS) infection when not recognized early. The overall mortality rate of bowel perforation is nearly 15% in shunted patients. Park et al. reviewed 50 cases in 2000, of which there were 10 deaths⁷¹².

The occurrence of a visceral perforation by the abdominal portion of a VPS is often a prolonged clinical phenomenon, which usually does not lead to acute peritonitis⁴⁷. Kin et al. described a case of a patient who was diagnosed with bowel perforation and chest/abdominal wall abscess 10 years after the VPS was implanted⁹. Acute visceral perforation may also occur due to catheter irritation leading to peritonitis, but this is the exception rather than the rule⁴⁷.⁷.⁹.

The most frequent finding in delayed bowel perforation by VPS catheters is extrusion of the distal catheter from the anus (45%)¹⁹. Ghritlaharey et al. observed 2.51% of VPS catheter perforation from the anus among all shunt surgeries in their series. The interval between shunt insertion to perforation of catheter from anus ranged from 2 to 20 months with an average of 6.1 months¹⁷. To Kanojia et al., the age at time of shunt extrusion ranged from 3 to 6 months with the average time interval of 3 months⁸. The present case stands for because the extrusion occurred long time after the shunt implantation (7 years).

Akyuz et al. proposed that the catheter tip adheres to the wall of viscera and a constant pressure of the abutting tip along with local inflammatory reaction leads to erosion of the visceral wall and entrance of the tip in the lumen. The peristaltic activity of gut carries it all the way down to the anus. Since the inflammation is usually a localized phenomenon, rarely are there any obvious signs of peritonitis⁵.

Ghritlaharey et al. also hypothesized the possible sequence of events for the occurrence of the spontaneous, silent bowel perforation with trans-anal VPS catheter protrusion as: (1) Due to the effect of gravity the peritoneal catheter may gravitate into the pelvis and penetrate into the colon/rectum; (2) Catheter migrate transmurally into the bowel lumen over a prolonged period of time, being surrounded by fibrous encasement at the enterotomy without subsequent peritoneal infection/peritonitis; (3) After eroding into the lumen of the bowel, the shunt usually propelled distally by the intestinal peristalsis⁵.

But even today, the exact pathogenesis of delayed bowel perforation by VP shunt catheter is not clear and it can be assumed that there more than one possible mechanism could work together in delayed bowel perforation by VPS catheter¹⁵. Among those, some mechanisms that have been suggested as potential contributing factors for bowel perforation are poor general condition of the patient with weakening of the intestinal wall¹ⁱ, the previous abdominal surgery³, distal catheter length¹⁶, presence of fibrosis around the distal tip¹⁶ and a stiff end of the shunt tube¹⁷.¹⁶.

Children are more susceptible to intestinal perforation because of their weak bowel musculature, especially those with myelomeningocele or congenital hydrocephalus, due to deficient innervations in the bowel wall¹⁴,¹⁵. Also, children have stronger peristaltic activity than adults, what would make them more susceptible to this kind of injury¹⁵. The majority of case reports and series describe more children with this complication than adults⁷,⁸. 60% of the patients from Ghritlaharey et al. series were children. According to Park et al., in their revision of 50 cases in literature, the mean age at perforation was 7.7 years, ranging from 1 month to 69 years.
Thirty-five patients were younger than 5 years and only 6 (12%) were older than 18 years. These authors stated that younger age was the main risk factor for bowel perforation. In the present case, the patient was an adult and did not have any known predisposing condition.

Other possible mechanisms were proposed by Brownlee et al., suggesting that allergic reaction to silicone, resulting in a foreign body–like reaction, might lead to adherence of shunt tubing to the intestinal wall with subsequent erosion into the lumen. Miserocchi et al. pointed out that the pathogenesis was most likely to be related with local infective adhesions of the bowel. Among the reasons for protrusion of the VP catheter from the anus, trauma during the operation should also be taken into consideration. A minor trauma to the bowel may cause a local inflammation which may lead the bowel wall to become more susceptible to the mechanical irritation by the catheter tip. Other mechanisms, such as a sharp tip of the shunting catheter, subclinical shunt infection, and increased protein content in the CSF have also been cited.

Non-enteric visceral perforation has also been sporadically reported and includes urinary bladder, vagina, gall bladder, stomach, scrotum, liver, uterus, and urethra. VPS catheter protrusion has also been reported from umbilicus, gastrostomy wound, healed neck wound, knee, and mouth. Metastases of a brain tumor via VPS to the abdomen have also been reported in literature.

In these cases of bowel perforation and anal extrusion of the catheter, the poor symptomatic scenario remains remarkable. In their series, Ghritlaharey et al. found none of their patients with features of peritoneal infection/peritonitis or intestinal perforation either at presentation or after shunt revision or removal. Shulhof et al., reported clinical peritonitis developed in 50% of their 12 patients series. It was initially present in two patients, but in four others it presented lately. Also, gram negative ventriculitis was present in five of 12 patients. In Park et al series, CSF infection was present in 46% of the cases, and the most common organism identified was Escherichia coli (48%), followed by Staphylococcus, Klebsiella, E. cloacae, and Proteus. These authors also stated that CSF infection was 3 times as common as peritonitis. Among the cases with infection, only 11% had both CSF infection and peritonitis. They noted as well that when a shunt catheter perforates the bowel, some mechanisms developed to prevent direct extension of infection from bowel to peritoneal cavity, for example, a fibrous encasement of the catheter along its intra-peritoneal course, which has been pathologically demonstrated in reported cases of bowel perforation, and coverage of the perforation site by the omentum. Since the majority of articles about this issue are case reports, it is difficult to precise the incidence of CNS infection secondary to bowel perforation, as well as the optimal treatment strategy in case of infection. Some authors listed warning signs for bowel perforation in shunted patients: meningitis or ventriculitis caused by an enteric microorganism (gram-negative bacilli), pneumocephalus, and abdominal symptoms, such as pain or prolonged unexplained diarrhea or fever.

Besides the obvious inspection when there is extrusion of the catheter through the anus, the imaging techniques for early detection of intra-abdominal complications secondary to VPS include radiography, sonography, CT and Magnetic Resonance Imaging (MRI). On radiography, the location of the shunt tip, displaced bowel gases, soft-tissue mass of pseudocyst, and intra-peritoneal free air can be seen. On sonography, the internal content, septa, and wall thickness of pseudocysts can be observed well. On contrast-enhanced CT or MRI, the peritoneal thickening, bowel-wall thickening and contrast enhancement, omento-mesentery infiltration, abscess, axial location of shunt tip and adjacent abnormal findings, and localized extraluminal air densities because of bowel perforation can be well evaluated. Among these techniques, however, CT may be more useful for the exact diagnosis of complicated intra-abdominal abnormalities. In their series, Chung et al. had one case (1.4%) of bowel perforation initially presented with subphrenic free air, and a subsequent abdomino-pelvic CT and exploratory laparotomy were performed showing transverse colon perforation by the catheter tip.

The management of this complication can be done in various ways. Among the different alternatives can be found laparotomy with revision of the peritoneal part of shunt, conventional exploratory laparotomy and repair of bowel perforation, endoscopic procedures, such as localization of enterotomy site and removal of shunt with the use of colonoscope, shunt removal and external ventricular drainage, use of antibiotics, followed lately by another form of CSF derivation. Ghritlaharey et al. performed a mini-laparotomy in 7 out of 10 of their patients, with revision of the peritoneal part of shunt, but shunt removal and delayed shunt revision was done only in 3 cases. Although the case report is...
some authors preferred removal of the entire shunt system, recently, a series of articles have mentioned more conservative alternatives of treatment. Chiang et al. reported the successful trans-anal treatment of the perforation, without the need of laparotomy. The authors stated that trans-anal repair of the colonic perforation after removal of the distal shunt may be considered an alternative to the usual laparotomy. In our case, we initially decided to proceed with laparotomy and adequate suture of the colon, but only reposition of the catheter that was not inside the colon lumen, without removal of the entire system. This was based on clinical scenario, on CSF initial analysis excluding CNS infection, neuroimaging, without any findings of infectious/malfunction of the VPS, and also blood exams that showed no signs of systemic infection. The decision for withdrawn the whole system was based on the new CSF culture, collected during the abdominal procedure that showed evidence of mixed infection.

Recently, in order to reduce the chance of perforation, some advancements had been made, such as the use of modern soft supple catheters made of silicone which may incite a lesser foreign body reaction.

In our opinion, early diagnosis, adequate clinical, radiological and biochemical evaluation, and prompt treatment are the key to successful treatment. Nevertheless, to these authors, the whole system should be always withdrawn, even when there is no clear evidence of infection in the preliminary exams. We also emphasize the necessity of establishment of a protocol of treatment to these patients.

In summary, bowel perforation with catheter extrusion through the anus is a relatively rare but potentially critical complication of VPS, much more common in children than in adults and usually occurs within the first year after implantation. It has been described, in its majority, through case reports and no standard treatment protocol exists for the treatment. It is important to early recognize this pathology and establish prompt treatment, in order to prevent CNS and abdominal infection.

REFERENCES


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