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Delayed brain abscess after bowel perforation by a subdural-peritoneal shunt

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Introduction

Among the many complications of Ventriculoperitoneal (VP) and subdural-peritoneal shunting, bowel perforation is infrequent, representing 0.1-0.7% of cases [1]. Recognition of this complication is important as the mortality rate is 15% secondary to intracranial and intra-abdominal infections [1,2]. Brain abscess is a rare manifestation of intracranial infection following spontaneous bowel perforation. To our knowledge, this is the seventh reported case of such an event, and the first case of delayed brain abscess formation after externalization and clamping of the catheter. The diagnosis of brain abscess, its radiologic features, and the possibility of delayed brain abscess formation are discussed.

Abstract

Placement of a subdural-peritoneal shunt is a common procedure in the treatment of external hydrocephalus. Abdominal complications such as bowel perforation are uncommon, and brain abscess formation following this event is even rarer. We describe the seventh case of spontaneous bowel perforation by a shunt catheter leading to brain abscess formation in a patient who presented with anal protrusion of the catheter. To our knowledge, this is the first case of delayed brain abscess formation occurring one month after catheter externalization. We discuss diagnosis, radiologic findings, and emphasize complete removal of the catheter, regardless of symptomatology, to avoid future brain abscess formation.

Case Report

Case reports are exempt from Institutional Review Board approval at our institution. A fourteen-month-old girl with a history of persistent macrocephaly, hypotonia, and developmental delay secondary to idiopathic external hydrocephalus underwent placement of a subdural-peritoneal shunt catheter with a one-way valve at eleven months of age. Intracranial pressure was normalat the time of insertion. She had no complications until twelve months of age when the distal end of the shunt catheter was seen protruding from her anus. She presented with mild irritability, but was afebrile and at her neurologic baseline.

At the time, the shunt catheter was externalized, cut at the



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level of the clavicle, and left the peritoneal portion in place. No Cerebrospinal Fluid (CSF) flow from the proximal shunt catheter was observed, and a sample for microbiologic evaluation was not obtained. An abdominal film showed the detached remaining catheter following the course of the descending colon with its distal end in the region of the rectum (Figure 1), suggestive of bowel erosion at the level of the descending and sigmoid colon. No free air was seen in the abdomen. A Computed Tomography (CT) of the head without contrast revealed no cerebral edema or mass lesion. No change in the size of the subdural space was noted when compared to prior imaging. The patient was treated with cefepime for three days and remained afebrile and clinically stable. As a result, lumbar puncture and surgical intervention were deferred. On hospital day #4, the shunt catheter passed spontaneously per rectum, and the remaining catheter was clamped at the level of the clavicle. Subsequently, the patient was discharged home.

One month later, the patient returned to the emergency department with fever, vomiting, and lethargy. A CT head without contrast revealed a large hypodense mass with surrounding vasogenic edema in the right frontal lobe suggestive of a brain abscess (Figure 2A). Further evaluation with contrast CT revealed a 4.0 cm by 5.5 cm lobulated, rim-enhancing mass in the right frontal lobe adjacent to the tip of the catheter (Figure 2B) consistent with a brain abscess. Additionally, a 2.1 cm by 1.3 cm lobulated abscess was located in the superior frontal gyrus. She underwent craniotomy for evacuation of the largest abscess, removal of the subdural-peritoneal shunt, and external drain placement. Surgical pathology confirmed the diagnosis of a right frontal brain abscess, and microbiology isolated Bacteroides fragilis and Klebsiella pneumoniae. The patient was subsequently treated with ceftriaxone and metronidazole.

Despite radiologic improvement on post-operative day #1 and #7, the patient had recurrent fevers. Follow-up Magnetic Resonance Imaging (MRI) on post-operative day #14 revealed a rim-enhancing lesion on post-contrast axial T1 imaging (Figure 3A) and high signal intensity on Diffusion Weighted Imaging (DWI) (Figure 3B) in the right frontal lobe. These findings were consistent with a brain abscess. Bilateral enhancement of the occipital horns of the lateral ventricular wall (Figure 3C) suggested ventriculitis. Axial T2 Fluid-Attenuated Inversion Recovery (FLAIR) imaging revealed increased signal intensity layering in the dependent portion of the lateral ventricles (Figure 3D) signifying intraventricular empyema. The patient was brought to the operating room for evacuation of the abscess and left frontal external ventricular drain placement, which led to stabilization of her condition. She was transferred to another institution on hospital day #32 for continued antibiotic therapy and placement of a subdural-peritoneal shunt.

Discussion

Idiopathic external hydrocephalus is a condition in which infants with rapidly enlarging head circumference are found to have a CT scan that shows widening of the subarachnoid space with mild or no ventricular dilation with an undetermined etiology. Subdural-peritoneal shunts are frequently used in the drainage of external hydrocephalus whereas VP shunts are used to treat communicating and non-communicating hydrocephalus. Associated complications include obstruction and infection with infection rates ranging from 3-29% and most often occur through the introduction of skin flora during initial shunt placement [3]. Twenty-five percent of all complications are abdominal symptoms such as pain or diarrhea with spontaneous bowel perforation occurring in only 0.1-0.7% of cases [1]. Perforation occurs more frequently in children, can occur weeks to years after shunt surgery, and most often involves the colon [4]. The presentation of bowel perforation varies, making its diagnosis clinically and radiologically difficult. In a review of forty-five cases of bowel perforation following VP shunt placement, 42% of cases were asymptomatic and 44% presented with a catheter at the anus. Other presentations included fever, abdominal symptoms, scalp necrosis, shunt dysfunction, meningitis, andseizures.2 Brain abscesses are much rarer and have been reported in a handful of case reports [5-9]. Despite its low incidence, a high index of suspicion for bowel perforation should be maintained because of possible intracranial and intra-abdominal infections with a mortality rate of 15% [1,2].

Direct visualization of the catheter protruding from the anus is an obvious sign of bowel perforation. Central Nervous System (CNS) infection by enteric organisms in a patient with CSF shunting is also highly suggestive of bowel perforation. In less obvious cases, nonspecific findings such as prolonged diarrhea or abdominal pain may be the only clues [10]. Serial abdominal radiographs can show the migration of the distal end of the catheter as it follows the colonic gas pattern [1]. Abdominal CT can identify the site of perforation [10] and can reveal an abscess or ascites [4].

The pathogenesis of bowel perforation by a shunt catheter is still unclear. Fibrosis of the distal tip may anchor the catheter within the peritoneal cavity. This leads to sustained pressure on the bowel wall eventually leading to its erosion and perforation. Recent abdominal surgery is also thought to be a predisposing factor due to inflammation of the bowel wall and decreased peristalsis [9,11].

Management of patients with bowel perforation ultimately involves removal of the catheter but should be individualized based on clinical presentation. In patients with signs of significant intra-abdominal infection, such as peritonitis or abdominal abscess, laparotomy or laparoscopy should be performed to repair the bowel [1,2,12]. Otherwise, a percutaneous1 or transanal approach [8,12] without bowel repair is appropriate as the chronic fibrous sheath is thought to seal the perforation site spontaneously with low risk of peritonitis [1,5].

Central nervous system infection can occur secondary to bowel perforation by a shunt catheter and results from the introduction of enteric flora into the distal end of the catheter. These organisms, mostly gram-negative bacteria such as Escherichia coli, travel in a retrograde fashion to reach the CNS.5 Infection may manifest as ventriculitis [2,7], meningitis [2], subdural empyema [5], or brain abscess [5-9]. We identified six previously reported cases of spontaneous bowel perforation by a shunt catheter leading to a brain abscess. Unique to our case is the development of a brain abscess one month following externalization and clamping of the proximal tip.

The suspected etiology of our patient's brain abscess was ascending infection of the intraperitoneal portion of the catheter. We propose that the infection extended to the portion of the catheter proximal to the externalized tip without reaching the brain because the patient had no signs and symptoms of CNS infection prior to externalization. The noncontrast CT at the time of catheter externalization showed no edema, mass effect, or sulcal effacement to suggest cerebritis. This proximal retained catheter served as the source of infection after the distal catheter connection to the perforated bowel was removed. Infection by isolated enteric bacteriaoccurred one month after this procedure supporting delayed brain abscess formation. Given this patient's course, it is reasonable to consider complete removal of the contaminated shunt in the absence of signs and symptoms of CNS dysfunction to avoid future infection.

Computed tomography and MRI are the imaging modalities of choice for the diagnosis of a brain abscess. A brain abscess typically begins as meningitis, progresses to cerebritis, and then develops into a pus-filled collection surrounded by a highly vascularized fibrous capsule [13]. On a CT head without contrast, a focal area of hypodensity with mass effect on the sulci or ventricle implying inflammation, edema, and abscess. With contrast, a rim-enhanced lesion with a smooth, regular thin-walled capsule appears representing an abscess [14]. In comparison, metastatic brain tumors often have irregular ring or nodular enhancement [14]. Other etiologies causing rimenhancing lesions such as Toxoplasmosis and glioblastoma multiforme must be correlated with patient history.

Magnetic resonance imaging has higher soft tissue resolution than CT and can better evaluate brain abscesses. Gadoliniumenhanced T1 imaging shows brain abscesses as rim-enhancing lesions with central hypointensity. On T2, a hypointense capsule surrounds a hyperintense central area [14]. On Diffusion Weighted Imaging (DWI), high signal intensity is seen because of decreased diffusion of water molecules through the lesion's viscous contents [15]. Although MRI has increased sensitivity and better differentiates rim-enhancing lesions, MRI involves a longer scan time and may require sedation, especially in young children. Given that our patient's history highly suggested a brain abscess, CT with contrast was appropriate in initially evaluating her condition.

Treatment for brain abscess following bowel perforation by a shunt catheter includes shunt removal, external drainage, and antibiotics [12]. Cultures of the evacuated fluid and tissue identify the causative pathogens and guide antimicrobial therapy. As in this case, the resolution of a brain abscess may be challenging and require repeated evacuation despite appropriate initial surgical and antimicrobial treatment. Treatment response is followed with repeat CT or MRI.

Because nearly 50% of patients with bowel perforation are asymptomatic, patients with VP and subdural-peritoneal shunts should be closely monitored. We emphasize shunt removal in cases of bowel perforation even without clinical or imaging evidence of CNS infection because delayed brain abscess formation can occur. Contrast CT and MRI with DWI can aid the early detection and diagnosis of cerebral infections and monitor the treatment outcome.

Figures



Figure 1: Abdominal x-ray in the anteroposterior supine view. The intraperitoneal portion of the shunt catheter (*white arrows*) is shown with its distal tip located in the area of the rectum (*black arrow*), presumably following perforation of the descending or sigmoid colon.



Figure 2: (Left) An axial CT without contrast shows several large areas of hypodensity (*white arrows*) in the right frontal lobe with surrounding vasogenic edema. **(Right)** An axial CT with contrast reveals a large rim-enhancing lesion (*white arrow*) in the right frontal lobe consistent with an abscess. In both images there is sulcal effacement and mass effect with a right to left midline shift. The shunt catheter (*black arrow*) is seen in the right frontal lobe.



Figure 3: Follow-up MRI on post-operative day #14. A brain abscess is shown in the right frontal lobe as **(A)** a rim-enhancing lesion (*white arrow*) on a post-contrast axial T1 image and **(B)** high signal intensity (*white arrow*) on diffusion weighted imaging (DWI) consistent with an abscess. **(C)** A post-contrast axial T1 image shows enhancement of the ventricular wall (*white arrow*) in the occipital horn of the left lateral ventricle suggestive of ventriculitis. **(D)** An axial T2 fluid-attenuated inversion recovery (FLAIR) image shows increased signal intensity layering dependently within the occipital horn of the left lateral ventricle (*white arrow*) suggestive of intraventricular empyema.

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