Shunt-Bronchial Fistula with Coughing Up and Swallowing of Cerebrospinal Fluid: Rare Complication of Ventriculopleural Shunt

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Key words
- Complication
- Fistula
- Hydrocephalus
- Persistent cough
- Recurrent pneumonia
- Ventriculoperitoneal shunt
- Ventriculopleural shunt

Abbreviations and Acronyms
CNS: Central nervous system
CSF: Cerebrospinal fluid
CT: Computed tomography
MRI: Magnetic resonance imaging
MRV: Magnetic resonance venography
VP: Ventriculoperitoneal

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BACKGROUND: **Erosion of the distal catheter into lung parenchyma is an extremely rare complication of ventriculopleural shunt placement.**

CASE DESCRIPTION: **We report a 51-year-old woman with a history of parasagittal meningioma invading the sagittal sinus who presented with recurrent pneumonia after placement of a ventriculopleural shunt. A nuclear study revealed accumulation of radiotracer material sequentially in the right hemithorax, trachea, mainstem bronchi, stomach, and bowel. The ventriculopleural shunt had eroded into the patient's lung parenchyma, with the effect of cerebrospinal fluid draining into the respiratory system and then being coughed up and swallowed into the gastrointestinal system.**

CONCLUSION: **Surgeons should be aware of the potential complication of a ventriculopleural shunt eroding through the lung parenchyma to cause a shunt-bronchial fistula with persistent coughing and recurrent pneumonias. Shuntogram nuclear imaging may be useful in the diagnosis of the complication.**

INTRODUCTION

Ventriculoperitoneal (VP) shunt is a neurosurgical procedure performed frequently for the treatment of hydrocephalus. It is indicated in congenital hydrocephalus, obstructive hydrocephalus, posttraumatic or posthemorrhagic hydrocephalus, and certain conditions that are refractory to medical management such as idiopathic intracranial hypertension. Although VP shunts are most commonly used, other locations such as the pleura (ventriculopleural shunt) and right atrium of the heart (ventriculoatrial shunt) may be used as distal termini for shunting of cerebrospinal fluid (CSF).We present the case report of a patient with a ventriculopleural shunt who presented with recurrent pneumonias and persistent cough who was found to have CSF spillage into lung parenchyma, and subsequently the trachea, stomach, and bowel, suggestive of erosion of the shunt tubing into the lung and then regurgitation of CSF from the respiratory tract into the gastrointestinal tract.

CASE REPORT

A 51-year-old woman with a medical history significant for posterior parasagittal meningioma with superior sagittal sinus invasion presented in November 2016 with headaches for neurosurgical evaluation at an outside institution. During her hospital stay, she was evaluated by ophthalmology and found to have papilledema; a lumbar puncture demonstrated elevated pressures. In January 2017, the decision was made to place a right frontal VP shunt. The meningioma was later treated with gamma knife radiation in February 2017. The VP shunt placement was complicated by an abdominal fluid collection, and the VP shunt was externalized and later replaced with a right ventriculopleural shunt in May 2017; all surgeries were performed at the outside institution. Operative records were not available. However, the patient did state that aside from the need for shunt externalization and reinternalization, there were no other complications or procedures (e.g., no pleural effusion or pneumothorax requiring intervention). She was never given any antibiotics.

After this, the patient established care at our institution in August 2017. During clinic visits, she continued to describe different symptoms, including persistent coughing, 2 episodes of pneumonia, headaches exacerbated by coughing, scapular pain, dizziness, syncope, and intermittent numbness and weakness. She stated that the coughing and bronchitis/pneumonia symptoms had started a few weeks after the ventriculopleural shunt placement. She was not taking acetazolamide or any medications for her headaches. Re-evaluation by her ophthalmologist revealed that her papilledema had improved. Magnetic resonance imaging (MRI) of the brain did not reveal signs of intracranial hypotension or shunt malfunction. Computed tomography (CT) of the chest did not show any pleural fluid collection, and the catheter

tip appeared to be in the pleural space without obvious migration into lung parenchyma.

A nuclear shunt study was performed in September 2017 to evaluate patency of the shunt. There was good spontaneous flow from the proximal shunt initially, suggesting no proximal obstruction; however, the CSF stopped draining spontaneously before an opening pressure could be obtained or enough sample could be collected for analysis without aspiration. A few hours after the shuntogram, the patient presented for evaluation upon suddenly experiencing severe headache, nausea, and vomiting. Her headache improved, although it did not resolve, with medical management including scheduled hydroxyzine, magnesium, and prochlorperazine (i.e., headache cocktail). Given that the patient had no neck pain or meningeal signs, there was low suspicion of meningitis. The patient started spiking fevers above 102°F and had leukocytosis of $15 \times 10^3/\mu L$ on arrival, so an infectious work-up was completed, and a chest x-ray showed a right middle lobe consolidation (Figure 1). She was taking vancomycin, piperacillin, and tazobactam, which was later transitioned to amoxicillin-clavulanate for the treatment of pneumonia. Ophthalmology evaluated the patient and did not find frank papilledema on examination. CT of the head demonstrated collapse of the right lateral ventricle over the catheter, and there was concern for overdrainage, similar to that observed 6 weeks before the procedure when the patient was seen in clinic (Figure 2). The result of CT of the chest, abdomen, and pelvis with contrast material was negative for effusion or obvious misplacement of the distal catheter of the ventriculopleural shunt, which was noted to course along a fissure with indeterminate penetration of the catheter tip into the lung parenchyma (Figure 3). The nuclear shuntogram (Figure 4) demonstrated extravasation of radiotracer material into the lung parenchyma, trachea, and stomach, concerning as a source of her recurrent pneumonia and persistent cough. It was concluded from the nuclear imaging study that the patient's ventriculopleural shunt had eroded into her lung to cause a shunt-bronchial fistula, which was the source of her infection and symptoms. As such, she appeared to have CSF drainage into the lung, which she subsequently coughed up and swallowed into the gastrointestinal tract.

At that time, it was recommended that the right ventriculopleural shunt be converted back to a right VP shunt; the patient was initially hesitant to undergo another surgery and required treatment for pneumonia. Six weeks later, she underwent removal of the pleural portion of the shunt with placement of a new distal peritoneal catheter in connection to the prior ventricular portion of the shunt, which demonstrated good flow of CSF. The valves were replaced intraoperatively, with the insertion of an ProGAV 2.0 valve (Miethke-Aesculap, Potsdam, Germany) set at 15 cm H_2O with a ProSA shunt assistant valve (Miethke-Aesculap) set at 22 cm H_2O in replacement of a nonprogrammable Delta valve (Medtronic Neurosurgery, Goleta, California, USA). We elected to change the valve to an MRI-compatible programmable valve with antisiphoning capabilities in case the patient needed adjustment because of over- or underdrainage. The abdominal portion of the surgery was performed laparoscopically with the assistance of general surgery because of the patient's prior abdominal surgeries and previous abdominal fluid collection. The patient's coughing resolved after removal of the pleural shunt.

The patient returned to the emergency room 8 days after surgery with abdominal pain and fever. CT of the abdomen and pelvis revealed multiloculated fluid collections extending throughout the pelvis, surrounding the uterus and also the midportion of the VP shunt catheter. CT of the brain demonstrated the VP shunt catheter in place with decreased ventricular size. The patient underwent shunt externalization at the clavicle and was given empiric antibiotics. CSF cultures from the shunt were positive for *Propionibacterium acnes*. The patient also underwent aspiration of the peritoneal and pelvic fluid by interventional radiology, for which cultures were negative. Infectious disease was consulted and recommended removal of the entire shunt. The externalized shunt drained 55 mL of CSF on the day of externalization, followed by 223 mL, 225 mL, and 77 mL on subsequent days before shunt removal. In November 2017 and 4 days after shunt externalization, the shunt was removed and a lumbar drain was placed. Cultures of the shunt tubing were similarly positive for *Propionibacterium acnes*. CSF cultures obtained intraoperatively and also 2 days postoperatively through the lumbar drain were both negative. Lumbar pressures were measured through the

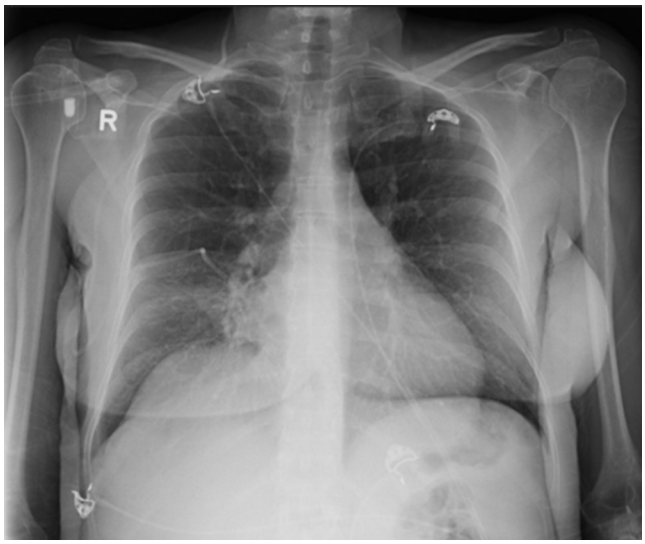

Figure 1. Chest x-ray demonstrating intact shunt tubing in the right side of the chest in addition to a focal opacity suggestive of atelectasis or consolidation in the right middle lobe.

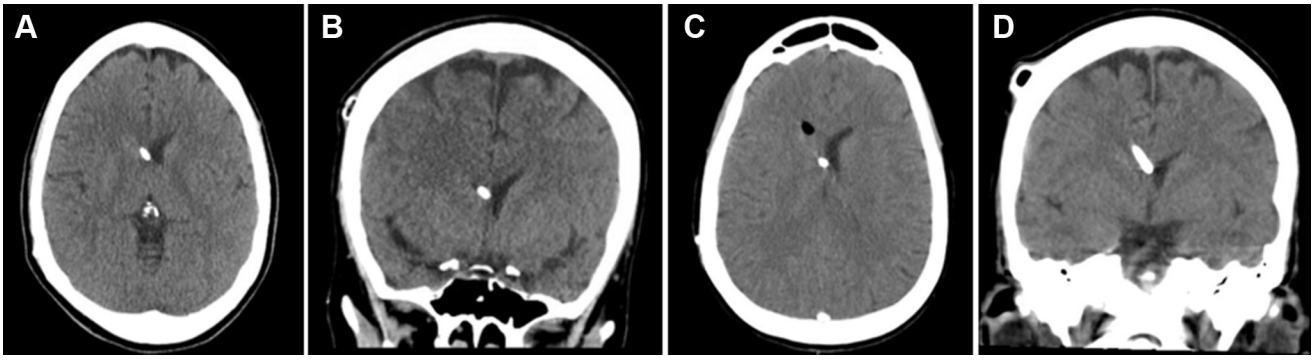

Figure 2. Computed tomographic axial (**A, C**) and coronal (**B, D**) images of the brain 6 weeks before the shuntogram (**A, B**) and same day after the shuntogram (**C, D**) demonstrate preprocedural and postprocedural collapse of the right ventricle with some pneumocephalus after the shuntogram.

lumbar drain with the patient in a supine position, without evidence of persistently elevated pressures above 20 mm Hg. MRI and magnetic resonance venography (MRV) of the brain demonstrated a 6-mm posterior interhemispheric left parasagittal meningioma causing mass effect (but not occlusion) on the posterior superior sagittal sinus, which was patent and draped along the left lateral aspect of the tumor (Figures 5 and 6). After discussion with the patient, given the findings of normal lumbar pressures and patent sagittal sinus, the decision was made to continue antibiotics for 2 weeks and return to the operating room after the course of antibiotics to obtain a lumbar opening pressure with the patient under anesthesia, and possible placement of a new VP shunt.

The patient was brought back to the operating room 2 weeks later. Lumbar puncture was performed, and pressure was measured over 5 minutes; it fluctuated between 21 and 22 cm of water, with CO_2 held constant at 35 and 5 cm of positive end-expiratory pressure applied to the ventilation. Given the patient's prior shunt complications and the pressure being on the higher end of normal, the decision was made to not place a VP shunt and instead to follow with serial lumbar punctures. A lumbar puncture in 1 month in December 2017 demonstrated opening pressure of 14 cm of water. The results of CSF studies obtained at that time were negative for infection, and ophthalmology evaluation was negative for papilledema. Additional lumbar punctures in January 2018, March 2018, December 2018, and August 2019 all demonstrated normal opening pressures <20 cm of water. The patient's most recent ophthalmologic evaluation in August 2019 showed continuing stability without papilledema, and her most recent MRI in July 2019 demonstrated a stable meningioma. There is currently no plan to re-implant a shunt system.

DISCUSSION RESUME HERE

Shunt malfunction is most frequently caused by shunt obstruction at some point along the system, which includes the proximal catheter, valve, and distal catheter.[1] Other complications include shunt fracture, disconnection at a junction, infection (which may produce obstruction), catheter migration, hardware erosion through surrounding tissues, overdrainage, seizures, extraneural metastases of central nervous system (CNS) tumors, ascites, bowel obstruction, pseudocyst formation, and fungating granuloma formation from an unknown silicone allergy.[2-4] Complications can also be due to

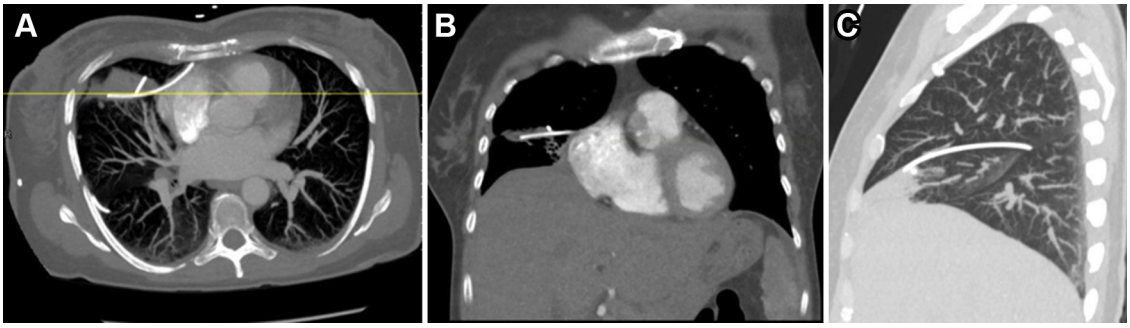

Figure 3. Axial (**A**), coronal (**B**), and sagittal (**C**) computed tomographic views of the chest, abdomen, and pelvis with contrast material demonstrating location of the tip of the ventriculopleural shunt. There was no pleural effusion, but atelectasis was noted in the right middle lobe adjacent to the ventriculoperitoneal catheter. The distal catheter was noted to course along a fissure with possible penetration of the catheter tip into the lung parenchyma.

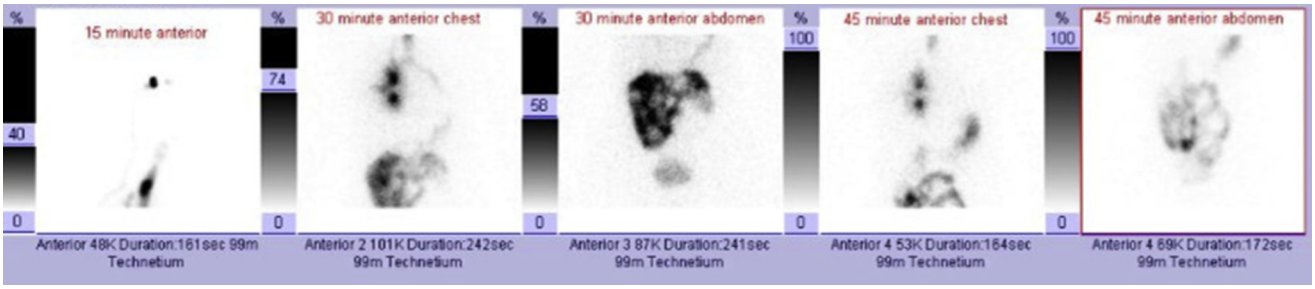

Figure 4. Nuclear shunt study findings compatible with a patent ventriculopleural shunt with spillage of radiotracer material into the right hemithorax consistent with location of the distal shunt catheter. There is accumulation of radiotracer material in the distal trachea and mainstem bronchi bilaterally, with later imaging demonstrating radiotracer material in the bowel and stomach. These findings raise concern for passage of radiotracer material into the airways.

surgeon error, such as intraparenchymal or intraventricular hemorrhage and malposition of either the ventricular catheter or the distal catheter. The distal ends of VP shunts have been shown to migrate into various adjacent regions, including the thorax, heart, pulmonary artery, liver, colon, bowel, stomach, gallbladder, urinary bladder, anus, breast, scrotum, and vagina.[5-16] Various fistulas may result from VP shunt migration: umbilical CSF fistulas, enterocutaneous fistulas, and bronchopleural fistulas.[9,17-19] Lumboperitoneal shunts may also migrate into the thorax and bronchus.[20] Ventriculoatrial shunting may be complicated by migration into the pericardium to cause cardiac tamponade or into the pulmonary system to create a bronchovenous fistula as late as 5 years after initial shunt implantation.[21,22] With regard to ventriculopleural shunts, complications described include pleural effusion, pneumothorax, hydrothorax, intrapleural pseudocysts, cardiac tamponade with heart failure, respiratory failure, or trapped lung.[23-32] A case report from 1962 described a patient with a ventriculopleural shunt who presented with severe headache in addition to long-standing irritating and constant desire to cough for several weeks. X-rays of the skull demonstrated air in the ventricular system, with a suspected diagnosis of bronchoventricular fistula, confirmed by the presence of spinal fluid trickling from the endotracheal tube during revision surgery.[33]

We present the second reported case in the literature and a very rare complication resulting from ventriculopleural shunting in which the distal shunt eroded through the lung parenchyma to create a fistula into the lung, resulting in recurrent pneumonia and persistent cough. Nuclear imaging demonstrated radioactive tracer material in the right hemithorax, trachea, mainstem bronchi, stomach, and bowel, suggesting that the CSF was entering the pulmonary system, being coughed up, and then swallowed into the gastrointestinal tract. Recurrent pneumonia has been reported as a complication of VP shunting, in which the shunt may erode through the diaphragm into the lungs.[7,15,16,34]

It has been speculated that low-grade infections or inflammation, coupled with the negative pressure of the pleural cavity, may facilitate the erosion of distal shunt catheters through structures such as the diaphragm and pleura.[7] The tubing may be misplaced during shunt implantation, or it may traumatically puncture an adjacent structure (e.g., the lung parenchyma) as the tubing gets stiffer over time. Our patient's CSF cultures after VP shunt explantation were positive for *Propionibacterium acnes*, which typically forms biofilm on prosthetic material. It is unclear whether the proximal catheter

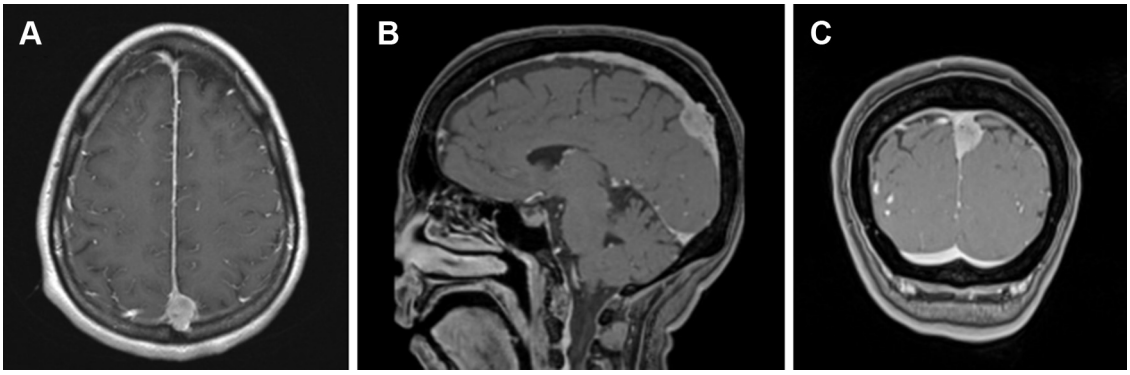

Figure 5. Axial (**A**), sagittal (**B**), and coronal (**C**) contrasted-MRI images demonstrate a 16-mm posterior interhemispheric left parasagittal meningioma causing mass effect on and extension into the superior sagittal sinus. MRI, magnetic resonance imaging.

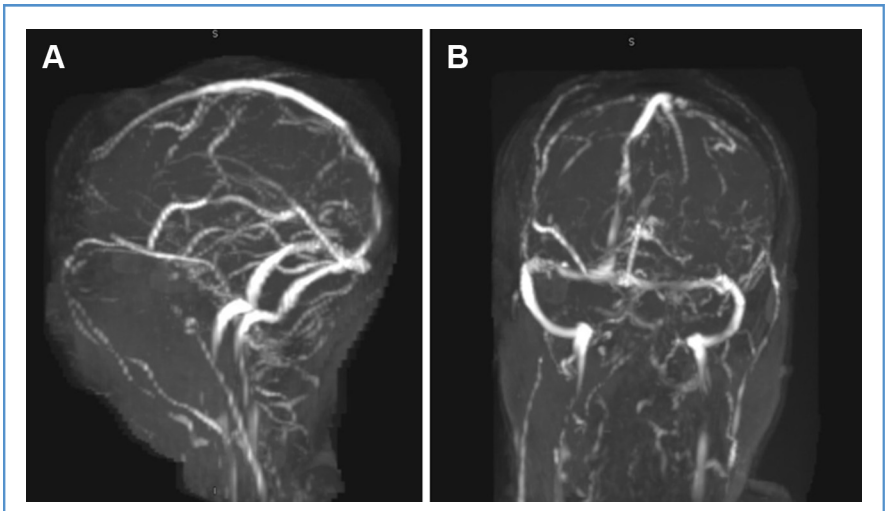

Figure 6. Sagittal (**A**) and coronal (**B**) MRV views demonstrate the posterior interhemispheric left parasagittal meningioma causing mass effect onto, but not occlusion of, the posterior superior sagittal sinus, which is patent and is draped along the left lateral aspect of the tumor. MRV, magnetic resonance venography.

and/or valve were replaced at the outside institution when the patient's VP shunt was replaced with a ventriculopleural shunt. She was never given long-term antibiotics, so it is possible that the shunt and/or the CSF was infected from the beginning, resulting in the subsequent pneumonia, erosion through the lung parenchyma, and abdominal fluid collections on 2 separate occasions. If CSF cultures were obtained during the initial shuntogram or during the conversion of the ventriculopleural shunt to a VP shunt, perhaps the infection would have been discovered sooner. CSF was unable to be obtained during the shuntogram, however, because there was collapse of the right ventricle around the catheter, perhaps resulting from overdrainage secondary to the negative pressure of the pleural cavity.

Distal shunt infections can manifest in various ways in addition to shunt malfunction: thoracic empyema or pleural effusions/pseudocysts in the setting of ventriculopleural shunts, abdominal pseudocysts or peritonitis with VP shunts, or bacteremia with ventriculoatrial shunts.[35-38] CSF should be sampled, and if possible the fluid collections at the distal site should be aspirated. If shunt infection is confirmed, the shunt should be removed, externalized, or removed, with placement of an external ventricular drain if the patient is shunt dependent.[39,40] The shunt may be replaced—perhaps to a different site—after appropriate antibiotic treatment and confirmation of clearance of the infection by means of serial CSF sampling or imaging of the distal site (e.g., to assess resolution of pseudocyst).[40] There is no consensus, however, on the appropriate management of a shunt in the setting of a distal site infection without a confirmed CSF infection. Whereas some case series describe the more conservative approach of shunt externalization or removal, Dalfino et al.,[41] for example, describe successful results in a series of patients with peritonitis in whom the distal VP catheter was left in place and simply managed with systemic antibiotics and local debridement when necessary.[37,41-45] In the case of our patient, the shunt was not externalized or removed at the time of presentation with pneumonia, was externalized after imaging showed multiloculated fluid collections in the abdomen/pelvis, and was later removed once a CSF infection was confirmed.

Additionally, we concede that it is unknown whether the distal end of the initial ventriculopleural shunt was placed appropriately, not into the lung parenchyma, given that the surgery was performed at an outside institution. Symptoms of cough and respiratory infections started about 2 weeks after ventriculopleural shunt placement and resolved after removal of the pleural component. Similarly, the size of the patient's ventricles before the initial shunt surgery and after presentation with the initial abdominal fluid collection is unknown, given that it occurred at an outside institution, although the patient did have papilledema and elevated lumbar puncture pressures, which were followed up over time. She was never given acetazolamide. After shunt externalization at our institution, the shunt drained more than 200 mL of CSF per day on the 2 full days it had been externalized, and thus we elected to remove it completely in accordance with infectious disease recommendations and to place a lumbar drain to measure pressures and assess the need for a shunt, inasmuch as we were unable to measure pressures through the externalized shunt, given the presence of a shunt valve.

This case report highlights the fact that unexplained respiratory symptoms (e.g., persistent cough) and repeated pneumonia in patient with ventriculopleural—or VP and lumboperitoneal shunts as evidenced by the literature review—should raise suspicion of fistula formation. Diagnosis will be based on a combination of clinical and radiographic findings. Bronchoscopy may be used to directly visualize the defect and assess for a possible need for repair.[20] Shuntograms with contrast material or radioactive isotope may be used to assess shunt patency.[46-50] These tests can be a useful adjunct in evaluating the flow characteristics of a shunt, assisting in diagnosing particular problems such as proximal obstruction (if no spontaneous flow occurs upon needle insertion) or distal obstruction (if no contrast material or tracer is found distally in the expected cavity at end of study). The shuntogram is not 100% sensitive and specific in diagnosing shunt malfunctions, however, so the overall clinical picture and additional testing all need to be taken into account.[47-49,51,52] On the basis of our experience and in accordance with other case reports, the defect in the lung parenchyma generally resolves spontaneously once the underlying cause is treated.[9] Additionally, this case illustrates that intracranial pressure arising from superior sagittal sinus occlusion—in this case secondary to a

tumor—may alleviate over time, as seen in this patient, who eventually did not require a VP shunt.

CONCLUSIONS

Recurrent pneumonia and persistent coughing in a patient with a ventriculopleural shunt can be caused by erosion of the distal shunt catheter into the lung parenchyma. A shuntogram may help reveal CSF passage from the shunt into the bronchi and trachea, which may then be coughed up and swallowed into the gastrointestinal system. Surgeons must be aware of this possible complication, so appropriate steps including shunt revision may be undertaken.

DECLARATION OF COMPETING INTEREST

The authors declare that the article content was composed in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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