

Delayed adult-onset recurrent symptomatic pleural effusion secondary to ventriculopleural shunt

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ABSTRACT

Background: Extracranial shunts reduce symptoms in patients with hydrocephalus by diverting cerebrospinal fluid (CSF) to an extravascular compartment. The most common procedure is ventriculoperitoneal shunting (VPS). However, certain patients with multiple shunt revisions, morbid obesity, or a history of abdominal surgeries may not be suitable VPS candidates; ventriculopleural shunting (VPLS) may be considered as an alternative route for CSF diversion when the peritoneal cavity is unsuitable. Pleural effusions, shunt obstruction, pneumothorax, and empyema can complicate the course of these shunts, though they are infrequent. Most reports of pleural effusions have been in young adults and children, particularly within three years of placement. We report the case of a 36-year-old woman who presented with recurrent symptomatic pleural effusion. The onset of symptoms was more than 25 years since insertion of VPLS. Initially managed with multiple therapeutic thoracentesis. Eventually patient underwent revision of shunt to VPS leading to resolution of symptoms.

Keywords: Ventriculopleural shunt, pleural effusion, thoracentesis

INTRODUCTION

Extracranial shunts alleviate symptoms in patients with hydrocephalus by diverting cerebrospinal fluid (CSF) to an extravascular compartment. The most common procedure is ventriculoperitoneal shunting (VPS), widely used regardless of the underlying cause of hydrocephalus. However, certain patients with multiple shunt revisions, morbid obesity, or a history of abdominal surgeries may not be suitable candidates for VPS. In such cases, ventriculopleural shunting (VPLS) may be considered as an alternative route for CSF diversion when the peritoneal cavity is unsuitable. Pleural effusions, shunt obstruction, pneumothorax, and empyema can complicate the course

of these shunts, though they are infrequent. Most reports of pleural effusions have been in young adults and children, particularly within three years of placement.^{1,2} Here we present a case of delayed onset of symptomatic pleural effusion due to VPLS in an adult.

CASE

A 36-year-old woman with a past medical history of recurrent pleural effusion presented with worsening shortness of breath. She initially presented with shortness of breath and cough a year prior to this admission. At that time, a chest X-ray revealed a moderate left pleural effusion with left lower lobe consolidation. Although a speech evaluation revealed normal swallowing, the patient was considered at high risk for aspiration due to a history of stroke, cognitive impairment, and episodes of choking. Consequently, she was treated for presumed aspiration pneumonia with antibiotics. However, her symptoms failed to improve,

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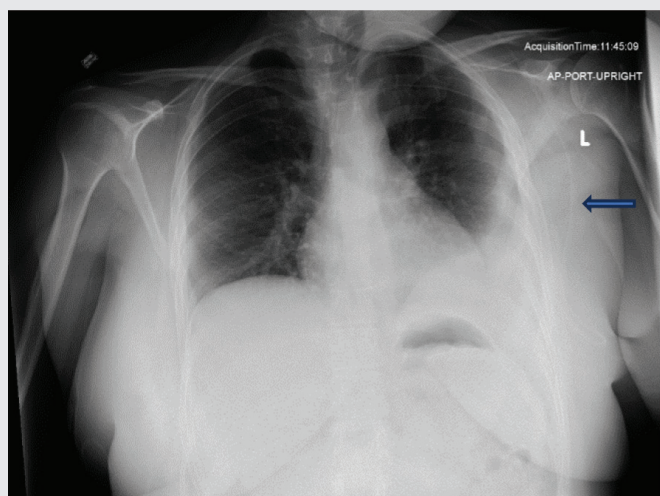


Figure 1. Chest X Ray revealing the tip of the catheter in the left pleural space (arrow), with moderate left pleural effusion.

and she subsequently underwent diagnostic and therapeutic ultrasound-guided (US) thoracentesis. Fluid analysis revealed transudative pleural effusion. Her symptoms resolved post-thoracentesis. Two months later, she returned with a similar presentation, and imaging revealed left-sided pleural effusion. Due to a lack of an alternate diagnosis, she was again treated on the lines of aspiration pneumonia, and therapeutic US-guided thoracentesis was performed.

Three months later, the patient presented again with symptomatic left-sided pleural effusion. Therapeutic US-guided thoracentesis was repeated, leading to symptom resolution. Interestingly, the radiologist noted the presence of shunt tubing on the chest X-ray done prior to discharge to establish a baseline for the pleural effusion (Figure 1). Computed tomogram showed predominant left pleural effusion with VPLS catheter in the left hemithorax (Figure 2).

A focused history obtained from the patient's mother revealed that, at the age of 2, the patient had undergone resection of an ependymoma, followed by five intracranial surgeries for debulking tumor regrowth. The patient unfortunately developed hydrocephalus, and a VP shunt was placed in early childhood. However, due to numerous shunt malfunctions and infections in the abdominal cavity, a VPLS had

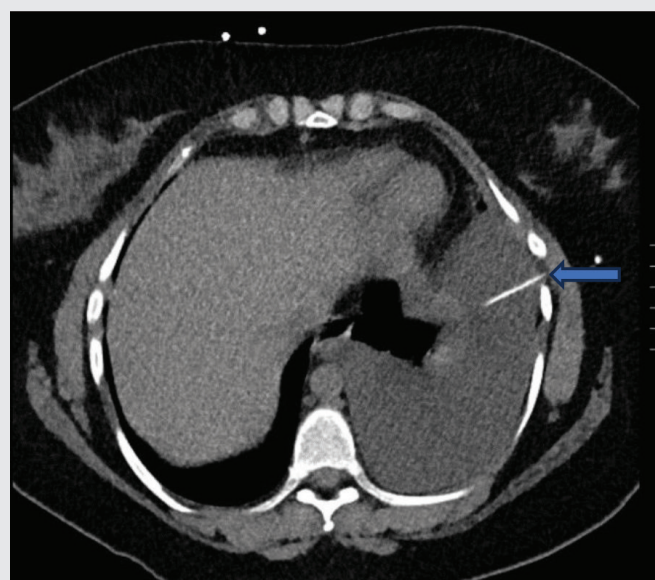


Figure 2. Computed tomogram showing predominant left pleural effusion with VPLS catheter in the left hemithorax (arrow).

to be placed over 25 years ago. Unfortunately, the failure of previous chest X-rays to identify the shunt, a lack of medical and surgical records from outside facilities, and a poor history given by the caregiver and family led to the failure to identify the presence of a VPLS for almost a year.

Neurosurgery was consulted due to concern for recurrent symptomatic pleural effusion as a complication of VPLS. Given the small but stable left lower pleural effusion and slit-like ventricles noted on the MRI, it was initially determined that the VPLS was functioning, and no acute intervention was required. However, two months after discharge, the patient presented again with symptomatic recurrent left-sided pleural effusion. Another therapeutic US-guided thoracentesis was performed.

The patient was started on acetazolamide as a conservative approach to decrease CSF production and possibly reduce pleural effusion volume. Eventually, the patient underwent definitive treatment with revision surgery to convert the left VPLS shunt to a VPS (Figure 3, Figure 4). A follow-up chest X-ray two months later did not reveal pleural effusion, and the patient continued to remain asymptomatic.

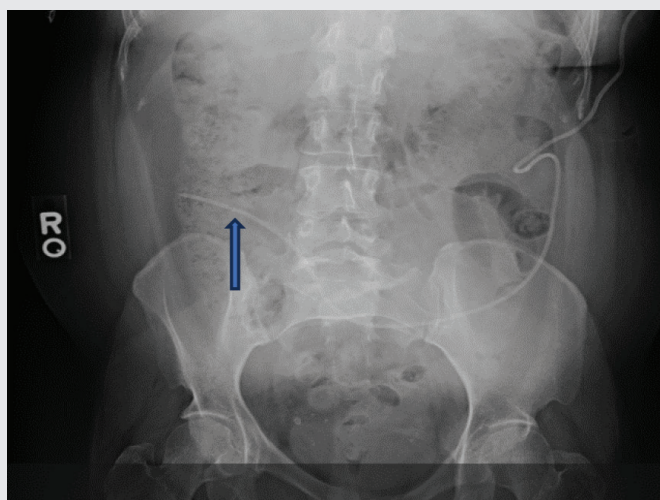


Figure 3. X Ray Abdomen after revision of shunt to VPS.

DISCUSSION

Ransohoff introduced VPLS as a management option for hydrocephalus in 1954. Although initially rewarding, long-term results were less satisfactory, with shunt obstruction or pleural effusions typically developing within three years.¹ Incorporating anti-siphon devices and valves into the shunt system has since been recommended to reduce the incidence of pleural effusions.³

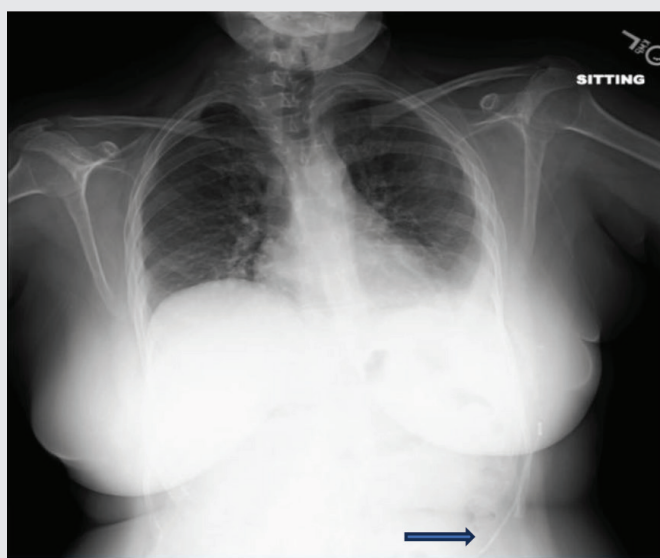


Figure 4. X Ray Chest after revision of shunt to VPS, left pleural effusion has resolved.

The mechanisms for the accumulation of pleural effusions with VPLS remain speculative. Two hypotheses may explain the development of hydrothorax as a complication of VPLS. First, impaired pleural absorptive capacity due to pleural damage secondary to prior infection and/or chronic exposure to CSF. Second, excessive drainage of CSF into the pleural space may affect this development. Other researchers have postulated that the negative pressure generated by the pleural space increases the risk of over-drainage. Evidence of CSF over-drainage can be seen on imaging as slit-like ventricles.⁴ Additionally, the presence of a shunt catheter in the pleural space may produce local irritant effects, inducing a chronic subclinical inflammatory response. Inflammation leads to increased pleural fluid production and impaired lymphatic flow, causing pleural fluid accumulation and lung collapse. This further reduces the pleural surface area, resulting in decreased net absorption of pleural fluid.⁷

A small, asymptomatic pleural effusion visible on chest X-ray indicates that the VPLS is functioning but does not necessarily imply dysfunction. Pleural fluid analysis usually reveals a clear, transudate-like fluid. Asymptomatic or minimally symptomatic effusions warrant close follow-up, while therapeutic thoracentesis can be used as a temporary measure for large symptomatic effusions. But revision of the shunt remains definitive treatment.⁸ Carrion et al. demonstrated that the administration of acetazolamide reduces CSF production, potentially increasing tolerance to VPLS and reducing respiratory symptoms.⁹

In our patient, VPLS was used as an alternative measure after the VP shunt course was complicated by shunt malfunctions and intra-abdominal infections. Interestingly, although the literature reveals that most cases of symptomatic pleural effusion due to VPLS occur within three years of placement, in our patient, the onset of symptoms was more than 25 years after the insertion of VPLS.

Due to its technical simplicity, high efficacy, and lower rate and severity of complications, VPS remains the procedure of choice for hydrocephalus. However, in cases of prior major abdominal surgery, a history of peritonitis, ascites, peritoneal dialysis, and failure of prior VPS, other alternatives for shunt placement, like

VPLS, may be considered.¹⁰ Although VPLS insertion is uncommon, it is occasionally used in patients with contraindications to VPS. Clinicians should be aware of the possibility of symptomatic pleural effusions complicating the course of these shunts, which may present even several years later. Shunt revision may be necessary in such cases for definitive treatment.

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Conflicts of interest: none

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