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Co-presentation of a subdural empyema and an infected ventriculoperitoneal shunt in an adult patient: A rare complication with review of literature

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Abstract

Background: The occurrence of a subdural empyema as a complication of a ventriculoperitoneal (VP) shunt infection is rare. Only three articles have been published on this topic. Moreover, the available literature only involves pediatric patients.

Case Description: The authors present a 38-year-old male with a preexisting right frontal subdural hygroma that developed into a subdural empyema in the presence of an infected right occipital VP shunt. A brief literature review is provided, and the pathogenesis is discussed.

Conclusion: This is the first known report regarding an adult patient with a subdural empyema and a VP shunt infection. Although a magnetic resonance imaging (MRI) brain is not typically ordered during diagnosis of a shunt infection, the authors advocate a low threshold to employ MRI brain to evaluate for other sources of infection, especially in an immunocompromised patient or in a patient with a history of a subdural hematoma or hygroma that can be easily overlook as being stable on computed tomography of head.

Key Words: Magnetic resonance imaging brain, subdural empyema, subdural hygroma, ventriculoperitoneal shunt

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INTRODUCTION

Subdural empyema is a critical neurosurgical condition that may stem from neurosurgical procedures, trauma, meningitis, sinusitis, or otogenic infection.[4,7] Reported mortality rates vary from 4.4% to 24%.[4,10,11,13] Devastating consequences include persistent seizures, hemiparesis, and stroke. Ventriculoperitoneal (VP) shunt placement is one of the most common procedures in neurosurgery. Moreover, the reported rates for shunt infections are relatively high (up to 10–15% in various studies).[5] However, their association with subdural empyema is rare. In the past, only 3 case reports have documented this association while another two patients are briefly mentioned in case series.[1,3,7,8,12] All reports concern the pediatric population. Herein, the authors describe the first adult patient who developed a subdural empyema from a preexisting hygroma shortly after placement of a VP shunt. Moreover, a literature review regarding this association is provided. The treatment of subdural

empyema often requires an urgent, if not emergent, operation for drainage. Undiagnosed, delayed treatment, or untreated subdural empyema has severe neurological consequences. A shunted patient was reported to have a subdural empyema undiagnosed and untreated for up to 9 years due to the lack of magnetic resonance imaging (MRI) and as a result, the patient suffered significant neurological injury.[8] The authors emphasize vigilance for this rare pathology in patients with existing VP shunts and advise a lower threshold for MRI. A prompt diagnosis of a subdural empyema offers the advantage of draining the infection and externalization of the infected shunt in the same operation, saving time, and money while giving the patient the best chance of recovery.

CASE REPORT

A 38-year-old male was consulted to rule out VP shunt infection. VP shunt was placed approximately 3 months prior to admission by an outside hospital (OSH). The patient presented with fever, emesis, and pancytopenia. Patient's history included a bilateral cerebellar mass resected by an OSH 3 months prior to admission. On postoperative day four, a right external ventricular drain (EVD) was placed due to hydrocephalus. Subsequently, the patient underwent right occipital VP shunt placement 10 days later. A follow-up computed tomography (CT) demonstrated interval development of right frontal subdural collection [Figure 1a-d]. Pathology of the resected tissue demonstrated medulloblastoma. A follow-up MRI brain 1-month later demonstrates the persistent right frontal subdural fluid consistent with hygroma [Figure 2a-d]. MRI of cervical, thoracic, and lumbar spines failed to demonstrate leptomeningeal metastases of medulloblastoma. The patient underwent radiation therapy with concurrent chemotherapy of vincristine, 2 months following surgery. Subsequent lumbar puncture did not show malignant cells.

Given the recent surgery, fever, and symptoms, there was serious concern for shunt infection. Cerebrospinal fluid (CSF) from shunt valve was obtained, which showed Gram-negative rods and Gram-positive cocci. MRI of brain with and without contrast revealed right frontal subdural fluid collection with ring enhancement and restricted diffusion, highly suspicious of subdural empyema [Figure 3a-d]. On examination, patient was awake and able to follow command in upper and lower extremities. Incision appeared clean and intact without signs of infection. The patient was taken to operating room, where the entire shunt system was removed and replaced with an EVD. In addition, a right frontal burr hole was placed over the area of subdural fluid accumulation and brisk drainage of purulent material was noted. Culture of this material grew out of *Escherichia coli*. The patient was immediately treated with broad-spectrum antibiotics. Patient's urine culture also grew out of *E. coli*. The patient was doing well and was back to his neurological baseline after surgery. Unfortunately, on postoperative day, three patients contracted severe pneumonia requiring intubation.

DISCUSSION

To our knowledge, there have been no prior reports regarding an adult patient with a subdural empyema

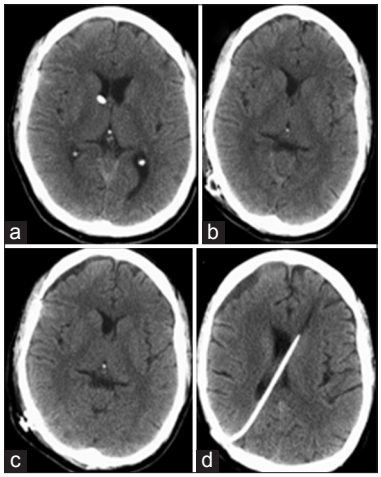

Figure 1: (a) Computed tomography (CT) head with right frontal external ventricular drain (EVD) in place; (b) CT head after right EVD removed and right occipital ventriculoperitoneal shunt placed with the present of a small right frontal hypodense extra-axial fluid; (c) CT head a few days later demonstrates slight interval enlargement of right frontal subdural fluid; (d) CT head on the day of admission, which is approximately 3 months postsurgery, for shunt infection redemonstrates moderate enlargement of right frontal subdural fluid

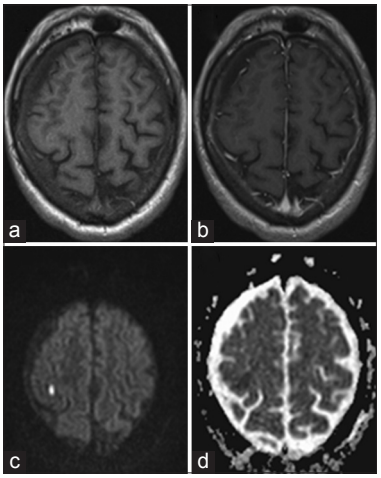

Figure 2: Magnetic resonance (MR) imaging was done 1-month prior to admission or 2 months postsurgery (a); MR T1 without contrast reveals right frontal subdural fluid (b); MRT1 with contrast reveals no enhancement of subdural fluid collection (c); Diffusion weighted imaging and (d); Apparent diffusion coefficient sequences failed to show restricted diffusion, suggesting a sterile subdural fluid collection that consisting with hygroma

after placement of a VP shunt [Table 1]. Though there are a limited number of reports, there may be a gender bias toward males. This may be due to the general incidence of subdural empyema, where there is a slight male preponderance.[7,9,10] Moreover, the patient with mucopolysaccharidosis was immunocompromised due to a recent stem cell transplantation; our patient had recently undergone chemotherapy and radiation with associated pancytopenia.

Subdural empyema can present with headaches, fevers, mental status deterioration, focal motor deficits, and seizure activity.[7] Its pathogenesis may depend on its etiology. A sinus infection may seed through valveless veins communicating between extracranial and intracranial structures; on the other hand, a neurosurgical procedure may introduce direct bacterial contamination of the subdural space or lead to secondary infection of a remote subdural effusion, both routes that ultimately lead to a subdural empyema.[3,8] Our patient had four neurosurgical procedures: The placement of a right frontal EVD, the posterior fossa craniotomy for resection of the medulloblastoma, the placement of a right occipital VP shunt, and the diagnostic lumbar puncture. After removal of the EVD and placement of the shunt, a minor subdural hygroma developed. This may have been a delayed by product of the EVD, and perpetuated by CSF diversion (from the shunt and/or lumbar puncture). An MRI brain with and without contrast 1-month after shunt placement confirmed the persistence of the subdural hygroma without signs of infection. Given how frequent shunt infections arise, compound with how rare subdural empyema occurs after EVD placement, the infection may have begun at the shunt, evolved to subclinical meningitis, and ultimately seeded the right frontal subdural hygroma.

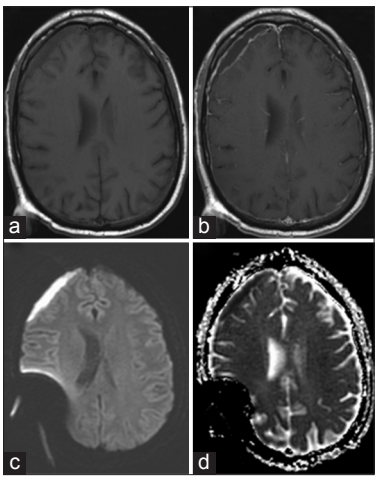

Figure 3: Magnetic resonance (MR) was done on the day of admission with the diagnosis of shunt infection or approximately 3 months postsurgery (a); MR T1 sequence without contrast reveals right frontal subdural fluid (b); MR T1 sequence with contrast reveals ring enhancement of subdural fluid collection (c); Diffusion-weighted imaging and (d); Apparent diffusion coefficient sequences show significant restricted diffusion, suggesting the development of a subdural empyema, which was not present a month prior

Another possible source of infection is the recent diagnostic lumbar puncture, which may cause meningitis and seed the hygroma and the shunt as well. A meta-analysis of 179 cases of meningitis after lumbar puncture revealed that approximately 9% were attributed to a diagnostic lumbar puncture; most of the other cases were linked to spinal anesthesia, where there is also injection of material into the spinal canal.[2] Postulated mechanisms for infection include contaminated instruments, poor technique, and aerosolized oropharyngeal secretions from procedural participants.[2] Moreover, there is a theoretical concern that under the circumstance of ongoing bacteremia, a lumbar puncture may induce meningitis.[6] Nevertheless, when Eng and Seligman[6] retrospectively evaluated 1089 bacteremic infants, the authors noted no significant difference regarding the incidence of meningitis for infants that received a lumbar puncture compared to those that did not. Our patient received a diagnostic lumbar puncture, where materials were not injected; however, he was immunocompromised, which increased his risk for postlumbar puncture meningitis. As an aside, he was not bacteremic when the lumbar puncture was performed.

A VP shunt infection is typically diagnosed after a workup that includes a CT head, a shunt series, and shunt reservoir tap. An abdominal ultrasound may also be obtained to rule out a pseudocyst. Once an infection is confirmed, the shunt is externalized, and the infection is

Table 1: Brief literature review with patient data

Source	Age	Gender	Etiology of hydrocephalus	Timing of presentation after shunt placement	Bacteria
Aliabadi *et al*.[1]	3 years old	Male	MPS syndrome	***	***
Dickerman *et al*.[3]	7.5 months old	Male	Congenital hydrocephalus	3 weeks	*Enterobacter* cloacae
French *et al*.[7]	***	***	Trauma	***	***
Kasliwal *et al*.[8]	17 years old	Female	Congenital hydrocephalus	9 years	***
Tahmouresie[12]	17 months old	Male	Congenital hydrocephalus	12 months	No growth of organisms
Current case report	38 years old	Male	Posterior fossa medulloblastoma	2 months	*Escherichia coli*

***No data. MPS: Mucopolysaccharidoses

treated appropriately. Typically, MRI brain is not ordered in the case of shunt infection. In fact, a shunted patient was reported to have a subdural empyema undiagnosed and untreated for up to 9 years due to the lack of MRI and as a result, the patient suffered significant neurological injury.[8] In this case, the radiologist reports a subacute on chronic right frontal subdural hematoma from the CT head on the day of admission [Figure 1d]. Therefore, relying on CT head alone, at least in this case, would miss the diagnosis of subdural empyema altogether, putting the patient at a higher risk of neurological injuries. We advocate a lower threshold for the use of MRI brain as part of the workup for shunt infection, especially if there is a history of a subdural hematoma or subdural hygroma, which is very commonly present in shunted patients.

CONCLUSION

Subdural empyema is an unusual complication of a VP shunt. There should be a high suspicion of this pathology in a patient with a history of a subdural hematoma or hygroma. Subdural empyema requires urgent, if not emergent, treatment to minimize neurological injuries. Consequently, there should be a low threshold to obtain MRI brain during workup for a shunt infection. A prompt diagnosis of a subdural empyema offers the advantage of draining the infection and externalization of the infected shunt in the same operation.

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