CASE REPORT

Ventriculoperitoneal shunt placement complicated by concurrent hydrothorax and hydrocele formation in a 4-year-old boy managed for congenital communicating hydrocephalus at a tertiary referral hospital in Mwanza, Tanzania

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Abstract

Hydrocephalus is among the leading causes of hospital admissions in the paediatric population of sub-Saharan Africa. The standard treatment of choice is the ventriculoperitoneal shunt; however, complications associated with ventriculoperitoneal shunt placement have been documented in the literature. Herein, we present a rare case of distal shunt migration with associated complications.

Keywords: hydrocephalus, ventriculoperitoneal shunt, hydrothorax, hydrocele, Tanzania

Introduction

Hydrocephalus is among the leading causes of hospital admissions for infants in sub-Saharan Africa.[1]-[4] The standard treatment of choice is the ventriculoperitoneal shunt (VPS), and the recent decade has seen the introduction of endoscopic third ventriculostomy as an efficient alternative in sub-Saharan Africa.[5] However, while VPS is the most common procedure conducted in the management of hydrocephalus, there is growing scientific evidence that complications of VPS may occur anywhere along the surgical tract, from the ventricular system to its blind destination within the peritoneal cavity.[6]-[12] Herein, we review a rare case of double shunting, which was complicated by distal migration; we discuss the course of management. Furthermore, consonant with the clinical picture, we briefly review similar cases reported in the literature.

Case presentation

We present the case of a 4-year-old boy who was admitted to our paediatric ward in September 2019 with a main complaint of severe respiratory distress for 4 days, which was associated with fevers and generalized body weakness. The patient was noted to have undergone a right-sided VPS procedure about 6 months earlier, in February 2019, due to a nonfunctional left-sided VPS that had been primarily placed in April 2018 for the treatment of congenital communicating hydrocephalus, diagnosed via computed tomography (CT) when the patient was 2 years old. Our review of the operative notes from the previous procedures revealed no documented difficulties during either of the VPS operations. Similarly, a review of the past medical history, including admission notes and oral history obtained from the parents prior to the procedures, showed no recorded evidence of hernia before shunting. The patient had been under post-VPS management at our institution for 8 months prior to this admission, with a reported history of intermittent, painless scrotal swelling persisting for more than 3 weeks. Upon admission, the child was tachypnoeic with chest indrawing and an oxygen saturation of 90% on room air. He was febrile with a body temperature of 37.9 °C, had a blood pressure of 100/70 mmHg, and had normal capillary refill. He was irritable but remained conscious and responsive to verbal commands. A thorough physical examination revealed that the patient was notably underweight for his age. He had bilateral ventriculoperitoneal shunts, both palpable from the occipitoparietal points and traceable along the trunk to the abdomen. The scrotum appeared swollen and was soft, mobile, retractable, and nontender.

Investigations

The initial full blood count (FBC) indicated leukocytosis with a predominance of neutrophils, and the malaria test returned negative. The electrolyte panel, liver function tests, kidney function tests, and urinalysis all fell within normal ranges. Analysis of cerebrospinal fluid (CSF) obtained from shunt tapping showed no abnormalities. A chest x-ray (Figure 1) displayed bilateral pleural effusions, notably more pronounced on the right side. Abdominopelvic ultrasonography detected a hydrocele and a potential right inguinal hernia, along with 2 VPS tubes present within the peritoneal cavity; the tip of the left-sided shunt was near the epigastric region. Abdominal ultrasonography was conducted to precisely locate the distal ends of both shunt catheters, and this revealed the left-sided tip in the lower peritoneal area and the right tip in the epigastric area, both demonstrating active CSF flow when the shunt valve was compressed

Differential diagnosis

Based on the above investigations, we established provisional diagnoses of possible shunt migration to the pleural cavity, bacterial pneumonia with respiratory distress syndrome, and a hydrocele secondary to an inguinal hernia. Differential diagnoses included shunt fracture, shunt overdrainage, and aspiration pneumonia, among other causes of respiratory distress.

Management

The patient received intravenous antipyretics and antibiotics and was placed on an oxygen mask, which led to improved oxygen saturation and resolved his fever. Following a consultation, the cardiothoracic team placed a chest drain for the right-sided pleural effusion, resulting in improved dyspnoea over 5 days. Beta-2 transferrin, a marker for CSF, was detected in the sampled pleural fluid, supporting the diagnosis of CSF hydrothorax. The neurosurgical team was consulted and found both shunt valves to be functional; however, the possibility of migration to the pleural space and shunt overdrainage was considered. The decision to remove the right-sided shunt was made because of the suspected overdrainage from both functioning shunts, a determination reached without the benefit of a preoperative CT scan of the head. Intraoperatively, the distal portion of the left-sided shunt, which was surgically truncated, was found near the pleura; however, no definite tract to the pleural cavity was identified. In the days following shunt removal, there was a notable reduction in the hydrocele and a progressive alleviation of the respiratory distress. Further CSF analysis did not detect bacterial growth, effectively ruling out bacterial ventriculitis. A CT scan of the brain, performed 5 days after external ventricular drainage, displayed ventriculomegaly indicative of increased intracranial pressure. This finding prompted the placement of a VPS, which was decided upon after a third consecutive negative CSF culture result.

Outcome and follow-up

A chest x-ray performed 7 days after the chest drain insertion showed complete resolution of the pleural effusion. On the first day following the VPS procedure, a control CT scan of the brain (Figure 2) confirmed the shunt's correct placement without evidence of increased intracranial pressure. Nonetheless, 3 days after the procedure, the patient's mother observed intermittent scrotal swelling, and the child's respiratory distress returned, even while he was receiving high-flow oxygen therapy. The patient's condition deteriorated,

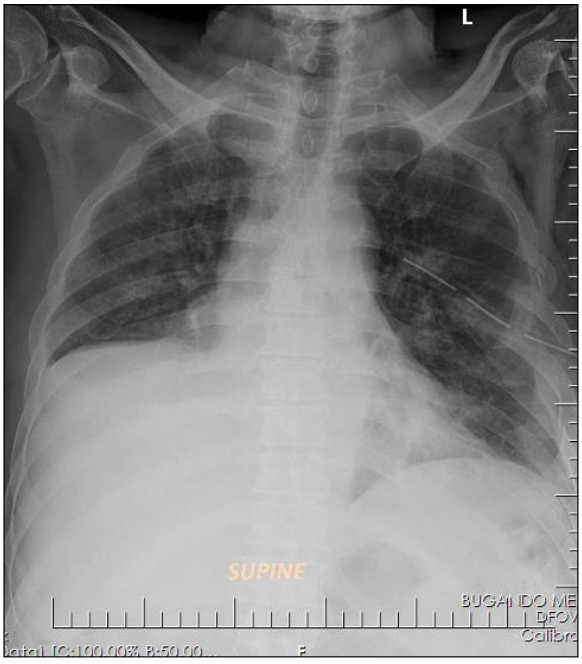

Figure 1. Chest x-ray showing a bilateral pleural effusion on admission

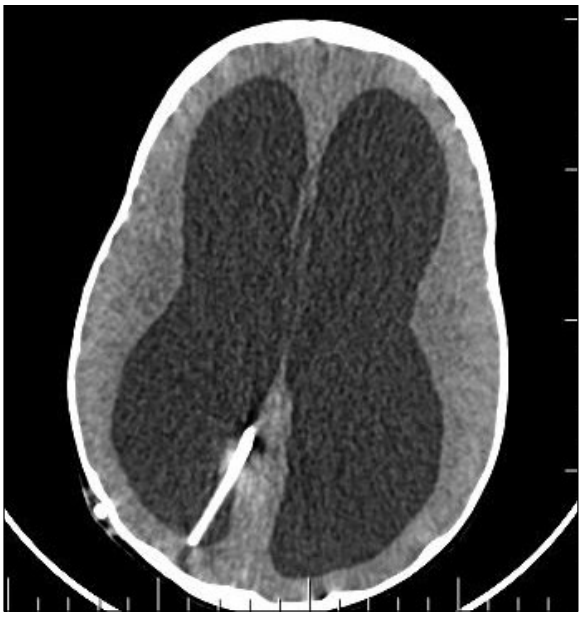

Figure 2. Noncontrast computed tomography scan showing the right ventricular ventriculoperitoneal shunt catheter in situ and ventriculomegaly on the first postoperative

marked by episodes of desaturation, high-grade fevers, and dyspnoea despite continuous antibiotic therapy and oxygen support. The patient died within 7 days following the VPS procedure, potentially due to severe pneumonia related to hydrothorax or aspiration pneumonia, compounded by prolonged immobilization. Shunt functionality was assessed and deemed operational, which discounted shunt failure as a cause of death. Despite aggressive treatment with high-dose intravenous antibiotics and mechanical ventilation in the intensive care unit, the patient succumbed to what was presumed to be severe pneumonia, characterized by severe respiratory distress. An autopsy was not conducted to conclusively determine the cause of death due to a lack of consent from the patient's family.

Discussion

Distal shunt migration is among the most commonly reported complications of VPS placement.[13]-[16] In this article, we describe a patient with 2 simultaneous complications of a VPS: left-sided distal migration into the pleural space and distal migration into the scrotum, leading to hydrothorax and hydrocele, respectively. Several authors have identified hydrothorax as a potential VPS complication, with varied management outcomes in both paediatric and adult patients.[17]-[21] Martin et al.[17] suggest that intra-abdominal migration of the catheter through the right costovertebral trigone of Bochdalek could be the mechanism behind hydrothorax formation.[17] In our case, we hypothesize that the truncated sharp end of the left-sided shunt inadvertently pierced the pleura, permitting the free flow of CSF into the pleural cavity and thus causing hydrothorax.

For the recurrent hydrocele observed in our patient, we propose that it was due to a congenital inguinal hernia that allowed positional leakage of CSF from the peritoneal cavity into the scrotum. A case report by Paterson and Ferch describes the effective resolution of a similar case of recurrent migration via shortening of the distal catheter.[22] Nawaz et al.[23] adopted a different treatment strategy for a patient with scrotal shunt migration, involving bilateral herniotomy, left-sided orchidopexy, and repositioning of the VPS into the peritoneal cavity in a 6-month-old with a recurring right hydrocele. Regrettably, in our case, the patient died from ongoing respiratory distress before additional surgery could be offered to address the hernia.

Conclusions

Hydrothorax, although infrequent, is a potentially fatal complication of VPS surgery and must be considered in any patient with a VPS presenting with severe respiratory distress. Prompt recognition and intervention of VPS migration, especially when it results in hydrothorax, are crucial to prevent dire outcomes. Additionally, recurrent hydrocele secondary to hernia formation typically necessitates surgical correction through herniotomy. The importance of a comprehensive, multidisciplinary team approach in managing such complex complications cannot be overstated.

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