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Case Report

Ventriculoperitoneal shunt-associated giant intraperitoneal cerebrospinal fluid pseudocysts: A case report and literature review

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ABSTRACT

Background: Ventriculoperitoneal (VP) shunt placement, a common treatment for hydrocephalus, can lead to various complications, including the formation of cerebrospinal fluid (CSF) pseudocysts within the peritoneal cavity. Although rare, occurring in 1-4.5% of cases, these pseudocysts pose significant clinical challenges due to their potential recurrence and the complexity of their management. The optimal management strategy depends on individual patient factors and the presence of infection.

Case Description: A 24-year-old woman presented with decreased consciousness, worsening headaches, and progressive abdominal enlargement. Imaging revealed a giant intraperitoneal cystic lesion, initially suspected to be a malignant ovarian cyst, but later identified as a CSF pseudocyst associated with the distal tip of a VP shunt placed 9 years earlier. The patient underwent urgent shunt revision, converting to a ventriculoatrial shunt. Postoperatively, her neurological status improved, and the abdominal mass resolved completely within 3 weeks. Follow-up over 1 year confirmed the absence of recurrence or neurological deficits.

Conclusion: VP shunt-associated intraperitoneal CSF pseudocysts, while rare, can develop long after shunt placement and present significant diagnostic and management challenges. Early recognition and appropriate surgical intervention are crucial to prevent complications. This case underscores the importance of individualized treatment approaches and diligent follow-up to ensure favorable outcomes, even in complex cases.

Keywords: Cerebrospinal fluid, Cyst, Neurosurgical procedures, Ventriculoperitoneal shunt

INTRODUCTION

Ventriculoperitoneal (VP) shunt placement can lead to various complications, including shunt obstruction, tip migration, infection, and issues with cerebrospinal fluid (CSF) drainage. [2] Among these, the formation of a CSF pseudocyst within the peritoneal cavity is a rare but serious complication, occurring in 1-4.5% of cases with a recurrence rate of up to 19.8%. [4,6,13,14] Although the precise pathophysiology of pseudocyst formation is not fully understood, contributing factors may include elevated CSF protein levels, peritoneal adhesions from previous surgeries, repeated shunt revisions, and impaired CSF absorption due to peritoneal inflammation. [2,7] In addition,

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inflammatory or allergic reactions to shunt materials, particularly silicone or ethylene oxide, may also play a role in pseudocyst development.[8] Management options range from surgical evacuation and shunt repositioning to alternative drainage techniques, depending on the presence of infection and the suitability of the peritoneal cavity for long-term shunt function.

In this report, we present the case of a 24-year-old woman diagnosed with a VP shunt-associated giant intraperitoneal CSF pseudocyst. We will detail the patient's medical history, diagnostic findings, management, postoperative care, and outcomes, followed by a discussion of this case in the context of existing literature.

CASE PRESENTATION

A 24-year-old woman presented to the emergency department with a complaint of decreased consciousness that had begun one day earlier. Previously, the patient had been fully alert, but she had suddenly become difficult to awaken in the morning and was disoriented when spoken to. She had been experiencing headaches for the past month, with the pain worsening over the previous week. In addition, the patient reported gradual abdominal enlargement over the past year, which had developed slowly and without pain. A week prior, she had undergone an abdominal computed tomography (CT) scan, which revealed a cystic lesion measuring approximately 23.9 \times 18.6 \times 28.9 cm, with solid components along the edges and minimal septations, extending from the pelvic cavity to the abdominal cavity [Figure 1a-c]. This lesion was displacing the stomach superiorly, the intestines anteriorly and posteriorly, and the uterus and bladder inferiorly, narrowing the ureters bilaterally at the medial to distal thirds and causing mild bilateral hydronephrosis. The distal tip of a VP shunt was visible within the cystic lesion. The patient was diagnosed with a suspected malignant ovarian cyst with peritoneal carcinomatosis, and surgery for cyst removal was planned by a gynecologist. However, the surgery was postponed as the patient was still considering it. There was no history of seizures, vomiting, visual disturbances, or bowel and bladder dysfunction. The patient had a VP shunt placed at the right Kocher's point 9 years earlier for hydrocephalus secondary to tuberculous meningitis, for which she had completed tuberculosis treatment. There was no family history of similar conditions.

On initial examination, the patient's Glasgow coma scale (GCS) score was Eye 3, Verbal 4, and Motor 6. Bilateral papilledema was observed, but there were no meningeal signs, neck stiffness, or other neurological abnormalities. A head CT scan revealed acute hydrocephalus, with the proximal tip of the shunt located in the third ventricle [Figure 2a and b]. We initially hypothesized that the hydrocephalus was due to distal shunt obstruction caused by compression from the intraperitoneal mass and malposition of the proximal shunt tip. The patient was advised to undergo revision surgery to convert the shunt system to a ventriculoatrial shunt.

The patient underwent a shunt system replacement with the insertion of a ventriculoatrial shunt on the same day. Cultures from the distal shunt tip showed no bacterial growth. On the 1st postoperative day, the patient's condition improved, and she regained full consciousness with a GCS score of 15 without any additional neurological deficits. The patient was monitored in the hospital for 5 days, and a follow-up CT scan confirmed proper positioning of the proximal shunt tip in the anterior horn of the right lateral ventricle [Figure 2c], allowing for safe discharge. At a follow-up visit 2 weeks postoperatively, the patient's abdominal mass had gradually decreased in size and completely resolved within 3 weeks after the procedure. Due to the patient residing in a remote area with limited access to healthcare facilities such as ultrasound or CT scans, further follow-up was conducted through telephone. Six months later, we conducted another remote follow-up. Clinically, the patient's abdomen appeared concave, with no visible or palpable mass on physical examination [Figure 3a and b]. An abdominal X-ray performed at a nearby healthcare facility revealed no signs of

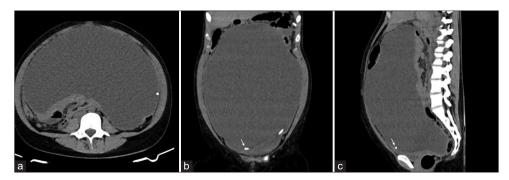


Figure 1: Non-contrast abdominal computed tomography scan showing an intraperitoneal cyst in the (a) axial, (b) coronal, and (c) sagittal views, with the distal shunt tip located at the base of the cyst (white arrow).

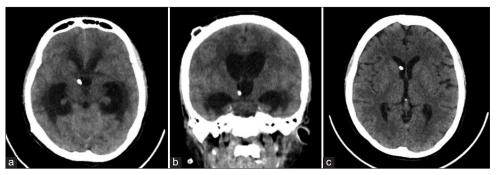


Figure 2: Non-contrast head computed tomography (CT) scans before and after shunt revision. (a and b) Pre-revision CT scans show the proximal shunt tip within the third ventricle in the (a) axial and (b) coronal views. (c) Post-revision CT scan shows the shunt catheter correctly positioned in the frontal horn of the right lateral ventricle.

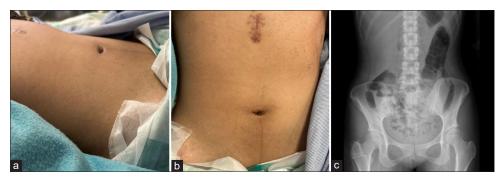


Figure 3: Patient examination during remote follow-up 6 months later. (a and b) Views of the patient's abdomen from (a) the front and (b) the side. (c) Abdominal X-ray in the anteroposterior view.

an intra-abdominal mass [Figure 3c]. Follow-up continued for 1 year postoperatively, during which the abdominal mass remained resolved without recurrence, and no neurological deficits or shunt malfunction were observed. The patient successfully resumed daily activities independently.

DISCUSSION

VP shunt placement is a common neurosurgical procedure used to treat hydrocephalus by diverting CSF from the ventricles to the peritoneal cavity. Although VP shunt procedures are generally safe and straightforward, complications such as shunt obstruction, shunt tip migration, infection, and under or over-drainage can occur. CSF pseudocyst formation is a rare complication, occurring in only 1–4.5% of cases, with a recurrence rate of up to 19.8%. [4,6,13,14]

While the pathophysiology of pseudocyst formation is not fully understood, several factors, including CSF protein levels, peritoneal adhesions from previous abdominal surgeries, repeated shunt revisions, and alterations in CSF absorption due to peritoneal inflammatory reactions, may contribute to this complication in both pediatric and adult patients. [1,2,6] Inflammatory and allergic reactions to the peritoneal catheter or CSF can also lead to pseudocyst formation, with 60% of cases associated with previous infections. [2] Allergic reactions to materials such as silicone or ethylene oxide used in shunts may also play a role.[8] The size of the pseudocyst is related to the risk of infection, with larger pseudocysts being mostly sterile, while smaller, loculated pseudocysts are more likely to be infected. [6] The organisms typically associated with these infections include Staphylococcus epidermidis, Staphylococcus aureus, and Propionibacterium acnes. [3,8] However, in our case, no bacteria were isolated from the distal shunt tip culture.

Pseudocysts are more commonly observed in children, with only 30 reported cases in adults.^[2] Children often present with symptoms related to shunt malfunction, such as headache, vomiting, and decreased consciousness. In contrast, adults typically present with abdominal symptoms, including pain, distension, palpable abdominal masses, nausea, vomiting, and anorexia; acute abdominal symptoms may also occur. [6,8] Only 30% of adults present with shunt malfunction symptoms. Our patient developed a pseudocyst 9 years after the initial shunt placement, likely due to a nonspecific local reaction to the shunt material in the peritoneum may have contributed to the development of the pseudocyst. In addition, the previous tuberculous meningitis may have exacerbated the inflammatory process in this patient. Pseudocyst complications were also previously published with similar symptoms to our case [Table 1]. Although the management of the cases was varied, most of the pseudocyst's cases were resolved.

Intraperitoneal pseudocysts can be detected through imaging modalities such as ultrasound and CT scan. These studies typically show a well-defined intraperitoneal fluid collection without septations, with the distal shunt tip located within or near the pseudocyst. [2,8] Missed diagnoses, such as in our case, can occur when the patient was initially diagnosed with a malignant ovarian cyst based on CT scan findings.[15] In cases of intracranial hypertension, a head CT scan may be performed to detect ventricular system enlargement and confirm shunt position.

Management of abdominal pseudocysts can vary. Surgical intervention with cyst evacuation and repositioning of the shunt catheter within the peritoneal cavity has a success rate of 70% and is the primary treatment option, especially in non-infectious cases. Other therapeutic options, such as external ventricular drainage, laparotomy, laparoscopy-assisted fluid drainage, and CT-guided needle aspiration with or without surgical evacuation, have also been reported in some cases. Ultrasound-guided aspiration may be performed to relieve acute symptoms while

	Authors	Year	Age	Clinical presentation	Management	Outcomes
1	Masoudi <i>et al.</i> ^[9]	2017	9 year	Abdominal distention, malaise, lethargic and slight fever	Laparotomy with cyst drainage and VP shunt	Recovery without symptom at 6 month follow up
2	Koide et al. ^[8]	2019	12 year	Disturbance of consciousness (GCS: E1V2M4), abdominal distention, headache	Extraventricular drainage and puncture cystic mass. Reinserted VP shunt	Recovery without complication
3	Raoof et al.[11]	2019	37 year	Abdominal distention and fatigue	Abdominal paracentesis with resected cyst mass and removed distal shunt catheters	Recovery without symptoms
4	Fatani <i>et al.</i> ^[6]	2020	7 year	Abdominal distention, headaches, and decreased oral intake	Transverse laparotomy with adhesiolysis followed by repositioned VP shunt	Recurrent after 8 months
5	Fatani <i>et al</i> . ^[6]	2020	5 year	Fever, vomiting, and abdominal distension	Laparoscopic exploration with adhesiolysis followed by VP shunt placement	Recovery until the last follow up at 18 months
6	Toubol et al.[16]	2020	37 year	Abdominal heaviness, vomiting and impairment of consciousness (GCS: 8)	Removed VP shunt, paracentesis abdominal cyst and ETV was performed	Successful remove of pseudocyst, recurrent abdominal pain and heaviness associated with mucinous cystadenoma
7	Jesus et al. ^[5]	2021	53 year	Fluid retention on the abdomen wall	Ventriculostomy drainage and catheter removal, VP shunt revised after 2 weeks later	Recovery until 2 months follow-up
8	Meyer et al.[10]	2021	4 year	Recurrent vomiting, abdominal distention	Revision VP shunt and evacuate CSF	Full clinical recovery
9	Wang <i>et al.</i> ^[17]	2021	68 year	Abdominal pain and distention	Laparoscopic cyst drainage with excision of the whole cyst, remove distal side of peritoneal shunt catheter.	Recovery without symptom
10	Risfandi <i>et al.</i> ^[12]	2022	62 year	Abdominal pain and lethargic	Exploratory laparotomy with excision pseudocyst and re-inserted peritoneal shunt catheter	Recovery without complication
11	Singh et al.[14]	2022	13 year	Abdominal distension, intermittent headache, and loss of appetite	Exploratory laparotomy with excision of pseudocyst and revision of abdominal end of VP shunt	Discharge on day 7

awaiting elective shunt revision. In cases of recurrent pseudocysts or shunt malfunction, as observed in our case, converting the shunt to a ventriculoatrial or ventriculopleural system may be a viable treatment option. If recurrent pseudocysts or shunt malfunctions occur, it may indicate that the peritoneal cavity is no longer a suitable long-term option for shunt placement, and shunt diversion to an alternative location should be considered. [2,6-8,14]

CONCLUSION

VP shunt-associated intraperitoneal CSF pseudocysts are rare but significant complications that can occur years after shunt placement. Factors such as elevated CSF protein levels, peritoneal adhesions, and inflammatory reactions to shunt materials contribute to their development. Accurate diagnosis is critical to avoid treatment delays. Management should be individualized, with alternative shunt systems considered in cases of recurrent pseudocysts or malfunction. This case demonstrates that, with appropriate intervention and follow-up, favorable outcomes are achievable without recurrence or neurological deficits.

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