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Review article

ABDOMINAL PSEUDOCYST DUE TO VENTRICULOPERITONEAL SHUNT: AN UNCOMMON COMPLICATION OF A COMMON NEUROSURGICAL PROCEDURE

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Abstract: For over a century, the ventriculoperitoneal shunt has been a standard neurosurgical procedure for treating hydrocephalus. However, this procedure is associated with a variety of complications. One uncommon but notable complication is the abdominal cerebrospinal fluid (CSF) pseudocyst. This pseudocyst is histologically characterized by a fibrous wall devoid of an epithelial lining, and its exact etiopathogenesis remains unclear. Patients with abdominal CSF pseudocysts often present with nonspecific symptoms, and treatment is tailored to each individual's clinical situation. This article reviews the epidemiology, etiopathogenesis, clinical characteristics, histology, imaging features, and available treatment options for abdominal CSF pseudocysts.

Keywords: Ventriculoperitoneal shunt, pseudocyst, neurosurgery, hydrocephalus, cerebrospinal fluid, shunt revision.

INTRODUCTION

Ventriculoperitoneal (VP) shunt surgery is a commonly performed neurosurgical procedure used to treat hydrocephalus. It takes advantage of the peritoneal surface's ability to absorb cerebrospinal fluid (CSF) (1). VP shunt complications are frequent, with 60% of shunts failing within ten years of placement, and up to 30% failing within the first year (2). Abdominal complications are particularly common and include fluid collections, peritonitis, gut perforation, shunt catheter displacement, fracture, migration, knot formation, and abscess formation (3, 4). Fluid collections secondary to VP shunt placement are relatively rare and typically present in two forms (5, 6): the accumulation of CSF (known as CSF ascites) and encapsulated fluid collections, referred to as abdominal pseudocysts (APCs). APCs, also known as 'CSFomas,' were first described by Harsh in 1954 (7). Their incidence is low, ranging from 0.33% to 6.6%, with a recurrence rate of 19.8% (8).

This article reviews the epidemiology, etiopathogenesis, clinical characteristics, histology, imaging features, and available treatment options for APCs.

Methods and materials

A comprehensive search was conducted in PubMed, Google Scholar, and ResearchGate using the following keywords: *'peritoneal cerebrospinal fluid pseudocyst,' 'pseudocyst after ventriculoperitoneal shunt,' 'abdominal pseudocyst,'* and *'complications of ventriculoperitoneal shunt.'* No specific timeframe was set for the literature search; however, articles published within the last two decades were prioritized.

Epidemiology

Abdominal pseudocysts (APCs) occur more commonly in children than in adults. The formation of an APC can take anywhere from three weeks to five years following the placement or revision of a VP shunt (6, 9). In some instances, formation has been reported to occur more than a decade later. For example, Wang HC et al. (10) described a 68-year-old woman who experienced increasing stomach pain and distention for four months. She had been treated for idiopathic normal pressure hydrocephalus with a VP shunt 15 years prior, and a 15-cm APC was detected upon evaluation (10).

Pathogenesis

Several hypotheses have been proposed to explain the pathophysiology of cerebrospinal fluid (CSF) pseudocysts, including elevated protein levels in CSF, a foreign body reaction to silicone, and alterations in CSF absorption caused by persistent subclinical inflammation or infection (9, 11).

In various studies, 17–80% of cases have been found to have a subclinical infection, with pathogens such as *Streptococcus*, *Staphylococcus aureus*, and *Staphylococcus epidermidis* identified from CSF cultures (12). However, repeated CSF cultures may appear sterile, and the infection can remain latent. In a review of 18 case reports, Ohba et al. (9) found that only 3 (16.7%) of the patients had *Staphylococcus epidermidis* cultured, while 15 (83.3%) had sterile CSF. Adults are reported to have a higher rate of infection than children (9).

Mobley et al. (13) suggested several risk factors for the development of APCs, including a history of necrotizing enterocolitis, previous shunt revisions, and prior abdominal surgery (excluding shunt revision). In contrast, Gmeiner et al. (14) found that the etiology of hydrocephalus, age at the first surgical procedure, and the type of first surgical procedure did not contribute to APC formation.

APCs may adhere to the parietal peritoneum or the serosal surface of abdominal viscera, or move freely within the peritoneal cavity. This explains why some APCs cause intestinal obstruction, while others undergo torsion.

APCs may also develop in relation to the liver, leading to hepatobiliary symptoms. Hepatic APCs can be classified as intra-axial or extra-axial. In intra-axial hepatic APCs, the tip of the VP shunt lodges within the liver parenchyma, forming a pseudocyst. In extra-axial hepatic APCs, the tip penetrates only Glisson's capsule, resulting in a hepatic subcapsular pseudocyst (15, 16).

Histopathology

An APC is characterized by a fibrous wall and the absence of an epithelial layer (Figure 1). This lack of epithelium differentiates APCs from true cysts (17,18,19). Other pathological features that may be observed in APCs include granulomatous tissue, acute inflammatory changes, lymphocytic infiltration, and an outer layer of fatty tissue of mesenteric origin (9).

Clinical Presentation

Symptoms of APCs are generally nonspecific. Patients may present with abdominal pain, distention, and a palpable lump, or may exhibit low-grade fever, backache, poor appetite, and shortness of breath (20). The pressure from the APC increases the VP shunt's resistance to CSF flow, which can lead to shunt dysfunction and raised intracranial pressure. This often manifests in children as headache, nausea, and vomiting (21).

APCs can grow to large sizes and, due to their mass effect, may obstruct the inferior vena cava, ureters, and intestines (22-25). Leung (22) reported a 14-year-old girl who presented with bilateral ankle edema as the sole symptom of a large non-infected APC. Imaging revealed obstruction of the inferior vena cava (IVC) and bilateral hydronephrosis caused by the APC. MPharm et al. (23) described a 12-year-old girl with bilateral lower limb pitting edema and abdominal distension; imaging showed compression of the IVC from a massive (20 × 18 × 8 cm) septate APC. Buyukyavuz et al. (26) reported a 3-year-old boy with a pseudocyst who presented with a hyponatremic seizure. Wang B et al. (27) reported a 19-year-old female with a VP shunt who presented with abdominal distension resembling that of a full-term pregnancy; the pseudocyst contained 12.7L of fluid.

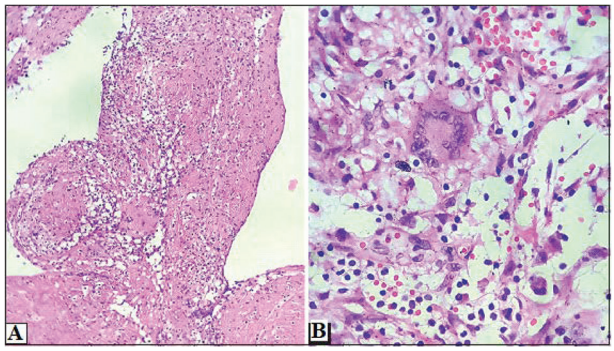

Figure 1. *Histopathology of CSF pseudocyst. (A) Photomicrograph showing a fibro collagenous cyst wall without definite lining epithelium (Hematoxylin and Eosin stain, 100×). (B) Photomicrograph showing cyst wall with many non-caseating epithelioid cell granulomas with Langhans giant cells (Hematoxylin and Eosin stain, 400×). Image credit: Shetty D, et al. Intriguing case of giant intra-abdominal pseudocyst: Diagnostic dilemma. Int J Health Sci (Qassim). 2020 Sep-Oct;14(5):58-60.Reused under the terms of the Creative Commons Attribution-Noncommercial-Share Alike 3.0 Unported*

Imaging studies

Ultrasound

Ultrasound is a rapid, non-invasive, and radiation-free imaging modality that can effectively diagnose APCs at a low cost (28, 29). On ultrasound, APCs appear as distinct hypoechoic or anechoic (black) fluid collections with well-defined hyperechoic (bright) margins. The tip of the VP shunt is often visualized within the pseudocyst as two hyperechoic (bright) lines (27, 28, 29), as shown in Figure 2. Chronic lesions may have multiple internal septations. Infected APCs typically exhibit debris and internal echoes (2). Larger lesions can exert pressure on adjacent organs (22-25).

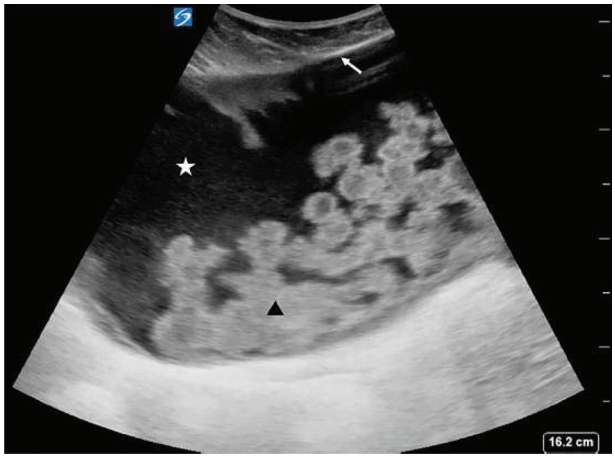

Figure 2. *Point-of-care ultrasound performed with a curvilinear probe in the right lower quadrant shows a large, anechoic (black) collection of cerebrospinal fluid (white star) encapsulated by a fibrous layer (white arrow) and containing echogenic debris and hyperechoic (white) septations (black arrowhead). Image credits: Guest et al. Abdominal Cerebrospinal fluid pseudocyst diagnosed with point-of-care Ultrasound. Clin Pract Cases Emerg Med. 2019 Jan 7;3(1):43-46. doi: 10.5811/cpcem.2018.11.40780. Reused under the terms of the Creative Commons Attribution-Noncommercial-Share Alike 3.0 Unported*

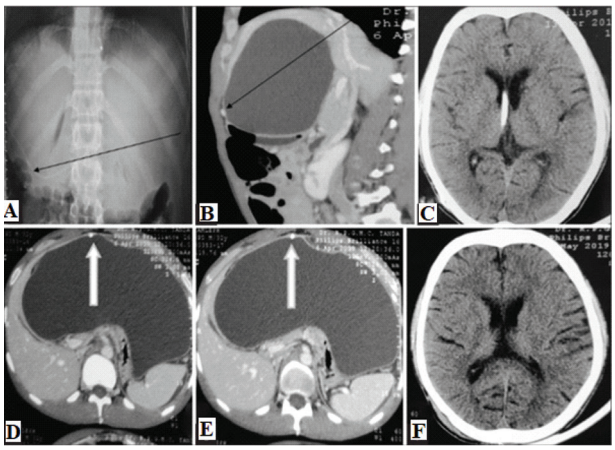

Figure 3. *(A)An X-ray showing the abdominal end of the shunt (black arrow) in the location of pseudocyst. (B, D, E) non-contrast computed tomography (NCCT) abdomen showing large pseudocyst abdomen with abdominal catheter lying along its anterior wall, as shown by black and bold white arrows. (C) NCCT head showing the ventricular end of the shunt. (E)NCCT head after removal of the shunt with no hydrocephalous. Image credits: Kumar M, et al. Central dilemma in CSF pseudocyst - A case series and review of literature. J Neurosci Rural Pract. 2022 Oct-Dec;13(4):753-758. doi: 10.25259/JNRP-2021-7-25. Reused under the terms of the Creative Commons Attribution-Non-Commercial-Share Alike 4.0 License*

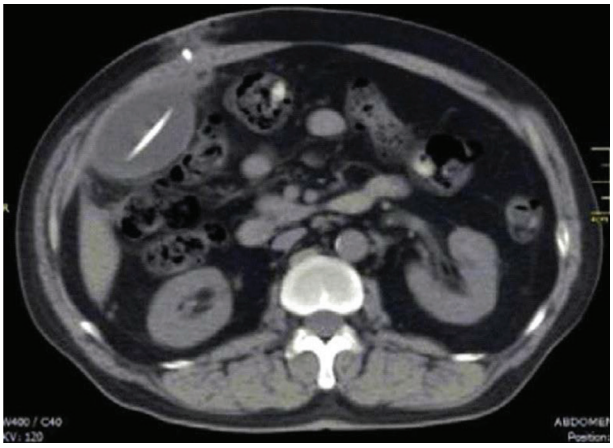

Figure 4. *Axial CT showing abdominal fluid collection adjacent to ventriculoperitoneal shunt catheter tip located on right abdomen. Image credits: Risfandi M, Celia C, Shen R. Abdominal wall pseudocyst as a complication of ventriculoperitoneal shunt insertion: a case report. Pan Afr Med J. 2022 Jan 10;41:23. doi: 10.11604/pamj.2022.41.23.29426. Reused under the terms of the Creative Commons Attribution International 4.0 License (*https://creativecommons.org/licenses/by/4.0/*)*

CT Scan

CT scans delineate a loculated cyst-like structure at the distal tip of the VP shunt, and the measurement of attenuation values helps characterize the water content (Figure 3 and 4). CT scans also assist in ruling out other etiologies, such as peritonitis, bowel obstruction, ascites, appendicitis, diverticulitis, abscess, and cystic abdominal lesions (20, 30).

Nuclear Medicine

A nuclear medicine shunt-o-gram can be used to identify APC. It reveals the normal passage of a radiotracer through the shunt tubing but without normal dispersion into the peritoneal cavity. Schmieder and Schraml (31) reported its successful use in a 73-year-old man who presented with forgetfulness, gait disturbance, and urinary incontinence. The shunt-o-gram showed normal antegrade flow through the patent tubing of the VP shunt, but with spillage into the peritoneal cavity without dispersion. CT further confirmed the diagnosis.

Plain X-ray Abdomen

A plain X-ray of the abdomen may reveal features of mass effect if the APC is large (Figure 3A). Similarly, it can identify signs of bowel obstruction or pneumoperitoneum, helping to rule out other differential diagnoses.

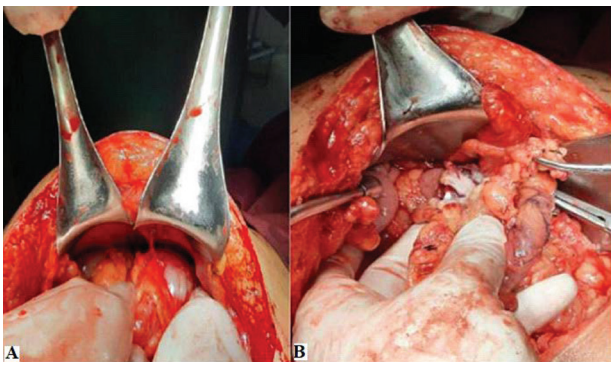

Figure 5. *Peritoneal cerebrospinal fluid pseudocyst as depicted on CT scan in Figure 3 : (A) pseudocyst before aspiration; (B) pseudocyst after aspiration of 750 ml of fluid, the distal shunt was identified. Image credits: Risfandi M, Celia C, Shen R. Abdominal wall pseudocyst as a complication of ventriculoperitoneal shunt insertion: a case report. Pan Afr Med J. 2022 Jan 10;41:23. doi: 10.11604/pamj.2022.41.23.29426. Reused under the terms of the Creative Commons Attribution International 4.0 License (*https://creativecommons.org/licenses/by/4.0/*)*

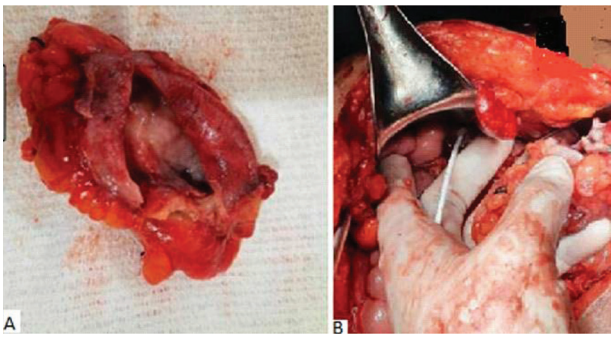

Figure 6. *Abdominal pseudocyst as depicted in Figures 3 &5. A) intraperitoneal cyst 10cm x 8cm x 7cm was excised; B) the distal side of the peritoneal shunt catheter was reinserted to the abdominal cavity. Image credits: Risfandi M, et al. Abdominal wall pseudocyst as a complication of ventriculoperitoneal shunt insertion: a case report. Pan Afr Med J. 2022 Jan 10; 41:23. doi: 10.11604/pamj.2022.41.23.29426. Reused under the terms of the Creative Commons Attribution International 4.0 License (*https://creativecommons.org/licenses/by/4.0/*)*

Differential Diagnosis

APC and CSF ascites are closely related differential diagnoses that can be difficult to distinguish. Clinically, shifting dullness is absent in pseudocysts and present in CSF ascites. Imaging technologies aid in making the final diagnosis. Cystic abdominal lesions such as pancreatic pseudocyst, cystic ovarian neoplasm, lymphocele, cystic teratoma, enteric duplication cyst, mesenteric cyst, and cystic mesothelioma are other differential diagnoses. A comprehensive evaluation employing cutting-edge imaging techniques and in-depth histopathology analysis is necessary for an accurate diagnosis.

Management

External drainage or surgical excision of APC (Figure 5 and 6) followed by shunt system reconstruction and repositioning of the abdominal catheter of the shunt are the major treatment options (1, 20, 21) mentioned in the literature. The approaches adopted for the surgical intervention are minimally invasive (laparoscopic) or laparotomy (Figure 6). There is no universally accepted therapeutic approach for CSF pseudocysts, and each patient's treatment should be tailored based on the specific clinical presentation. Following external drainage and cessation of CSF inflow, the absence of secretory epithelium results in collapse and gradual disappearance of APCs.

If the peritoneal cavity is unsuitable due to adhesions or infection in shunt-dependent patients, the

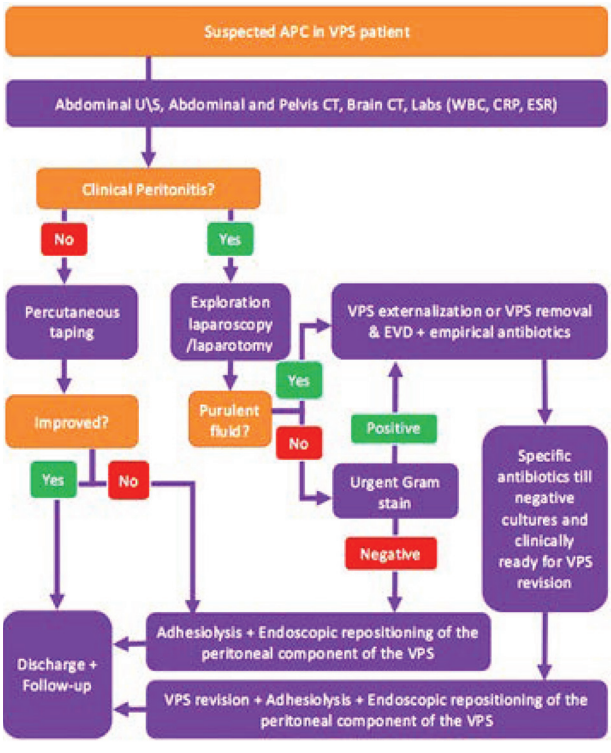

Figure 7. *An algorithm that summarizes the approach for patient with abdominal CSF pseudocyst. Image credits: Fatani GM, Bustangi NM, Kamal JS, Sabbagh AJ. Ventriculoperitoneal shunt-associated abdominal cerebrospinal fluid pseudocysts and the role of laparoscopy and a proposed management algorithm in its treatment. A report of 2 cases. Neurosciences (Riyadh). 2020 Aug;25(4):320-326. doi: 10.17712/nsj.2020.4.20200053. Reused under the terms of the Creative Commons Attribution-NonCommercial License (CC BY-NC)*

abdominal end of the VP shunt may be externalized and placed at an alternate site on the same or contralateral side of the abdomen after eradication of infection. Ventriculo-atrial (VA) shunt, ventriculo-pleural (VPL) shunt, or endoscopic third ventriculostomy are the other available options if the abdomen is not suitable. In patients with shunt independence, shunt removal may be undertaken safely.

Various algorithms have been proposed in the literature to suggest management strategies (20, 33). An easy-to-apply algorithm was proposed by Fatani et al. (32), as depicted in Figure 7.

CONCLUSION

Ventriculoperitoneal shunt placement poses a lifetime risk of complications, necessitating careful monitoring. Every healthcare provider should be aware of abdominal pseudocysts and consider them when making a differential diagnosis in patients with VP shunts who present with vague abdominal symptoms. Treatment is tailored according to clinical circumstances, and the management strategy continues to evolve.

Abbreviations

VPS: Ventriculoperitoneal shunt
CSF: Cerebrospinal fluid
APC: Abdominal pseudocyst
IVC: Inferior vena cava
CT: Computed tomography scan
NCCT: Non-contrast computed tomography scan
VA: Ventriculoarterial
VPL: Ventriculopleural
EVD: External ventricular drain

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Author Contributions: Each author took ownership of the project and contributed to the article's composition, critical revision, and data analysis.

Note: Artificial intelligence was not utilized as a tool in this study.

Sažetak

ABDOMINALNA PSEUDOCISTA KOD VENTRIKULOPERITONEALNOG ŠANTA: RETKA KOMPLIKACIJA ČESTE NEUROHIRURŠKE PROCEDURE

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Ventrikuloperitonealni šant je standardna neurohirurška procedura za lečenje hidrocefalusa već više od jednog veka. Iako je efikasna, procedura je povezana s različitim komplikacijama, među kojima je abdominalna pseudocista retka, ali klinički relevantna. Ova pseudocista histološki se karakteriše fibroznom kapsulom bez epitela, dok njena tačna etiopatogeneza ostaje nejasna. Klinička slika je nespecifična, što otežava dijagnozu, a terapijski pristup se prilagođava individualnim karakteristikama pacijenta. U ovom radu se razmatraju epidemiologija, etiopatogeneza, kliničke karakteristike, histološki nalazi, radiološke osobine i dostupne terapijske opcije za ove abdominalne pseudociste.

Ključne reči: ventrikuloperitonealni šant, pseudocista, neurohirurgija, hidrocefalus, cerebrospinalna tečnost, revizija šanta.

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