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Case Report

Radiology Section

Intra-axial Hepatic Cerebrospinal Fluid Pseudocyst: An Unusual Complication of Ventriculoperitoneal Shunt Placement

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ABSTRACT

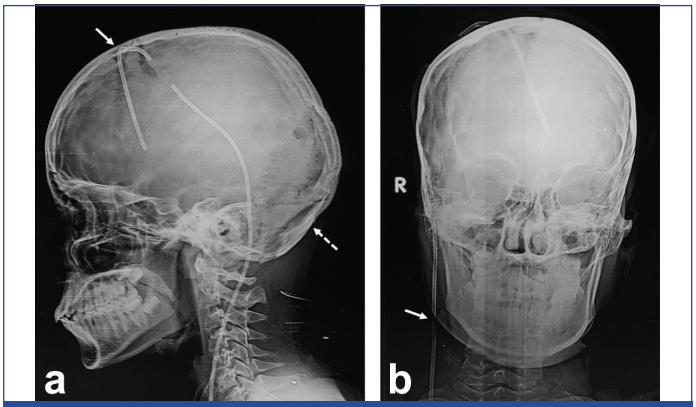
Intra-abdominal Cerebrospinal Fluid (CSF) pseudocysts are a well-known and potentially serious complication of Ventriculoperitoneal (VP) shunt placement. These cysts are often found within the peritoneal cavity or along the serosal surface of solid organs. Here, we present an unusual case of an intrahepatic CSF pseudocyst in a 14-year-old female patient with a VP shunt placed three years ago, following the excision of a cerebellar pilocytic astrocytoma. The child presented with complaints of right hypochondriac pain and tenderness for one month. On ultrasound of the abdomen, the right lobe of the liver showed a complex anechoic cystic collection with multiple thick internal septations. A Contrast-enhanced Computed Tomography (CECT) examination revealed a well-defined hypodense collection in segments IV and V of the liver with a VP shunt tube within. Based on clinical and imaging features, differentials of intra-axial intrahepatic CSF pseudocyst, complex hepatic cyst, peri-shunt hemorrhagic cyst, and biliary cystadenoma were considered. Aspiration of the cyst revealed clear, colourless, non purulent, non sanguineous fluid which tested positive for beta-2-transferrin, confirming the collection to be a CSF pseudocyst. An elective laparoscopic cyst excision with shunt revision was performed with no recurrence on follow-up. The present case highlights the uncommon occurrence of intrahepatic CSF pseudocysts post-VP shunt, necessitating meticulous diagnostic evaluation. Successful laparoscopic excision and shunt revision underscore the efficacy of tailored interventions. This contribution enhances the understanding of atypical complications in VP shunt recipients, guiding clinicians and radiologists in both diagnosis and management.

Keywords: Aspiration, Beta-2-transferrin, Cyst excision, Intrahepatic collection

CASE REPORT

A 14-year-old female patient presented to the Outpatient Department (OPD) with complaints of non colicky dull aching pain in the right hypochondrium for the past month. The pain was progressive in nature, with no aggravating or relieving factors, and there was no history of vomiting or altered bowel habits.

The patient had previously undergone surgery for cerebellar pilocytic astrocytoma with obstructive hydrocephalus, which was treated by suboccipital craniotomy, near-total excision of the tumour, and VP shunt placement three years ago [Table/Fig-1a,b].



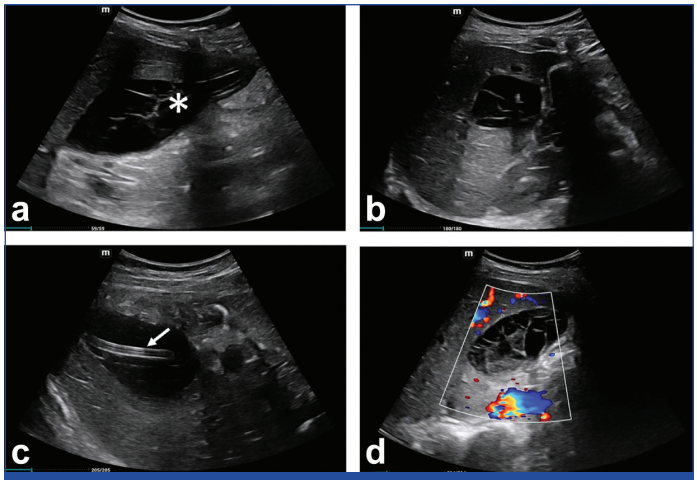
[Table/Fig-1]: A 14-year-old female child presented with complaints of abdominal pain and tenderness diagnosed with intra-axial hepatic CSF pseudocyst. Shows a) lateral; and b) anteroposterior radiograph of the skull showing Ventriculoperitoneal shunt (VP) entering the cranial cavity through the burr hole defect (arrow) and craniotomy defect (dotted arrow) involving the occipital bone. The VP shunt tube is seen coursing along the right side of head and neck (arrow).

Upon clinical examination, the patient's vital parameters were normal, and there were no signs of pallor, icterus, clubbing, cyanosis, lymphadenopathy, or pedal oedema. There were no clinical signs of chronic liver disease. Examination of the abdomen showed mild hepatomegaly (the right lobe of the liver was palpable 2 cm below the costal margin). Additionally, a palpable slightly tender soft to firm mass in the right hypochondriac region, which was found to be moving with respiration.

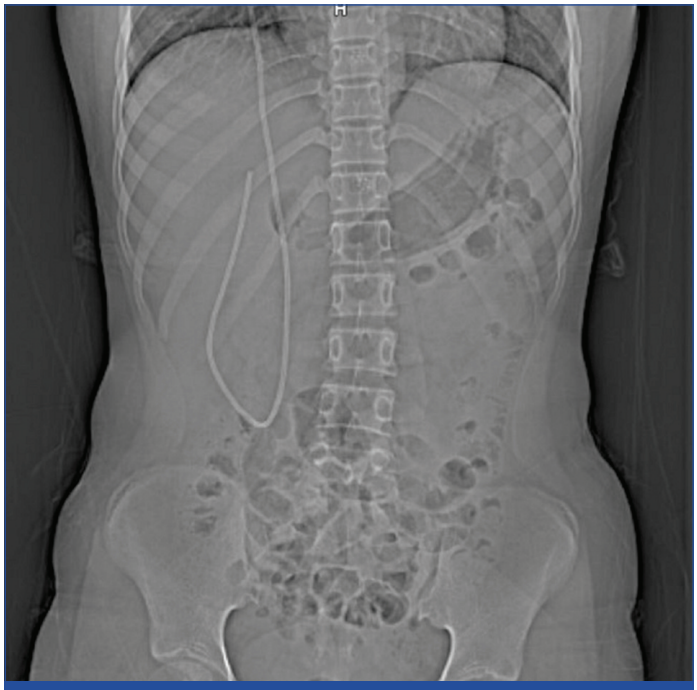
Haematological values at admission showed mild anaemia (Haemoglobin 9.5 gm/dL (12-15 gm/dL)) and mild leucocytosis, White Blood Cell (WBC): 11×10^3/microliters ($4\text{-}10\times10^3$ microliters). Liver enzyme levels were marginally elevated Serum Aspartate Transaminase (AST): 52 U/L, Serum Alanine Aminotransferase (ALT): 58 U/L, Serum Alkaline Phosphatase (ALP): 415. Renal function tests were within normal limits. Based on the clinical features and lab parameters, the clinicians considered the possibilities of hepatic hydatid cyst, hepatic abscess, and acute hepatitis. Consequently, Immunoglobulin G antiamoebic and antiechinococcus granulosus antibody tests were performed, which were found to be negative.

The patient was then referred for an ultrasound of the abdomen. Both grayscale and colour Doppler ultrasound examination were performed, revealing a well-defined anechoic complex cystic collection within the parenchyma of the right lobe of the liver with multiple thick and thin internal septations. The colour Doppler showed no internal vascularity [Table/Fig-2a,b,d]. Additionally, a hyperechoic tubular structure within the cystic collection [Table/Fig-2c] corresponded to the tip of the VP shunt.

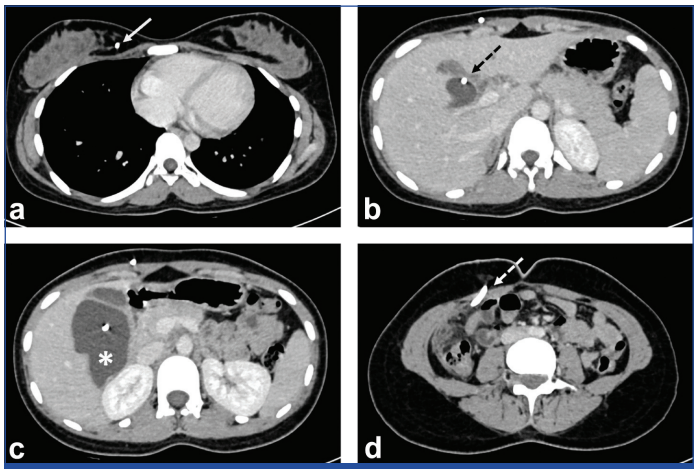
Based on the ultrasound findings, the differentials of intrahepatic Cerebrospinal Fluid (CSF) pseudocyst, biliary cystadenoma, and hepatic abscess were considered. As a result, the patient was referred for a computed tomography of the abdomen to confirm the findings. The topogram showed the trajectory of the VP shunt tubing with its tip positioned in the right upper quadrant of the abdomen [Table/Fig-3]. CECT revealed a well-defined hypodense intrahepatic collection of fluid attenuation (+5 to 10 HU) in segments IV and V of the liver with the VP shunt tube within. The VP shunt tube had curved back within the collection and coursed cranially with its tip located at the hepatic hilum [Table/Fig-4a-d]. A rim of hepatic



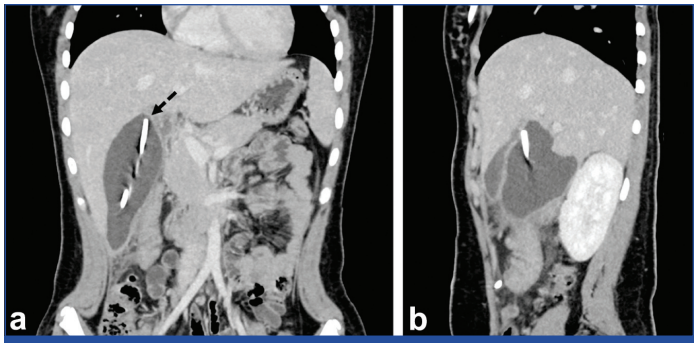
[Table/Fig-2]: a) Gray scale longitudinal ultrasound image of liver showing a well-defined anechoic intraparenchymal cystic collection in segments IV and V of liver (asterisk); b) Ultrasound image along the transverse axis; c) Ultrasound image showing linear hyperechoic tubular structure within the collection suggestive of Ventriculoperitoneal shunt (arrow); d) Doppler showing no internal flow.



[Table/Fig-3]: Topogram demonstrate Ventriculoperitoneal (VP) shunt tubing coursing along the right chest wall and abdominal wall and curled within the right iliac fossa then coursing up with its tip in right upper quadrant of the abdomen in the expected location of the cystic collection.

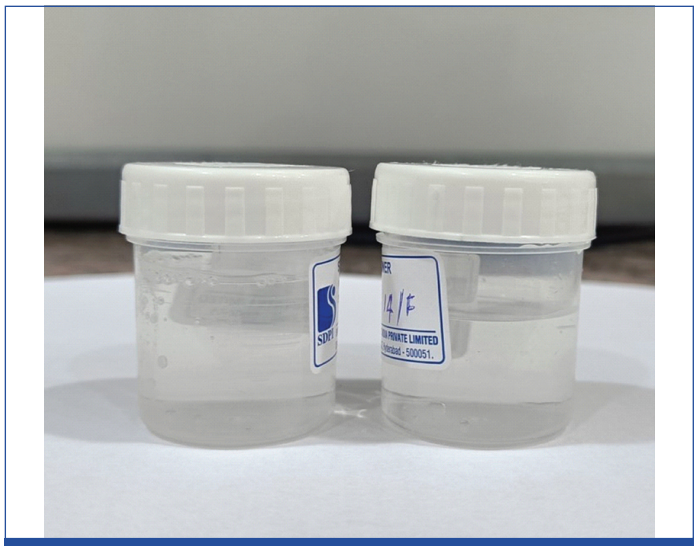


[Table/Fig-4]: Contrast enhanced axial CT images of lower chest and upper abdomen showing well-defined hypodense intraparenchymal collection (asterisk) involving segment IV, V of liver measuring 7.4×5.2×11.6 cm (APXTRXCC) with Ventriculoperitoneal (VP) shunt coursing along the subcutaneous plane of right anterior chest and abdominal wall (white arrow), entering the peritoneal cavity (white dotted arrow), coursing back with its tip in the intra-parenchymal liver collection (black dotted arrow).



[Table/Fig-5]: Reformatted CECT abdomen a) coronal and b) sagittal section show well-defined hypodense intraparenchymal liver collection with demonstration of the tip of the Ventriculoperitoneal (VP) shunt within the collection (black dotted arrow).

parenchyma could be identified along the periphery of the collection [Table/Fig-5a-b]. No serpiginous membranes, daughter cysts, or peripheral rim of calcifications were seen in the collection, and there was no perilesional oedema. Based on the imaging features, the differentials of intrahepatic CSF pseudocyst, resolving perishunt haemorrhagic cyst, and biliary cystadenoma were considered.



[Table/Fig-6]: Fine needle aspirate of the pseudocyst showing clear, colourless fluid.

To confirm the diagnosis, an ultrasound-guided aspiration of the collection was performed under strict aseptic precautions using a 23 G quincke-type point needle. Around 35 mL of clear, colourless, non purulent, non sanguineous fluid [Table/Fig-6] was aspirated.

The fluid was sent for biochemical and microbiological analysis, which showed Red Blood Cells (RBCs) (20 cells/mm^3), WBCs (Total leukocyte count: 80 cells/mm^3, polymorphs: 10%, lymphocytes: 90%), glucose: 47 mg/dL, protein: 0.3 g/dL (Albumin: 0.2 g/dL), Lactate Dehydrogenase (LDH): 83 IU/L, ADA: 1.4 U/L, and amylase: 1 U/L. The microbiological analysis revealed no growth of organisms after aerobic as well as anaerobic incubation with scanty polymorphs on gram staining. Auramine and rhodamine staining showed no acid-fast bacilli. The sample showed positivity for beta-2-transferrin, confirming it to be CSF.

Three weeks later, the patient underwent laparoscopic drainage and cyst excision with shunt revision while receiving broad-spectrum antibiotics. Intraoperatively, a thin hepatic tissue layer covering the cyst was found, which contained colourless, non purulent fluid, confirming the diagnosis. The patient was discharged after two days, with oral broad-spectrum antibiotics for a period of two weeks. A six-month follow-up abdominal ultrasonography revealed complete resolution of the collection with no evidence of residual/recurrence.

DISCUSSION

The VP shunting, which uses the peritoneal cavity to absorb CSF, was first used in 1905. Since then, VP shunting has become one

of the most prevalent procedures used to treat hydrocephalus [1]. In obstructive hydrocephalus, positioning the proximal end of the VP shunt is based on the surgeon's preference. The distal end is advanced along the subcutaneous plane with a shunt passer to the abdomen, where an incision is used to access the peritoneal cavity where the tip is placed [2].

According to published literature, 5 to 47% of patients with VP shunts experience a variety of abdominal complications [3]. These complications include peritonitis, migration of the catheter into the colon, CSF pseudocyst, CSF ascites, extrusion of the catheter into an inguinal hernia, and intestinal obstruction [4]. Intra-abdominal pseudocysts are an uncommon complication and occur with a frequency of 1 to 3% [5] and are known to occur more frequently in children [6]. Risk factors for the formation of a CSF pseudocyst include multiple shunt revisions, infection, peritonitis, and adhesions [7]. The proposed mechanism for the development of a hepatic CSF pseudocyst is inflammation triggered by prolonged irritation of the hepatic surface by the shunt catheter. However, in the present case, we could not isolate any organism from the aspirate.

Most CSF pseudocysts form within the peritoneal cavity. They are thought to be due to a low-grade infection around the shunt followed by omental wrapping [8]. After formation, they may move freely or may adhere to the parietal peritoneum, bowel loops, or the serosal surface of solid organs [9]. However, CSF pseudocysts forming within a solid organ are rare, and very few cases have been described in the medical literature [10], where solid organs like the liver and spleen have been involved [3].

Hepatic CSF pseudocysts are very rare and are classified as intra-axial and extra-axial hepatic CSF pseudocysts. A comprehensive literature review revealed a total of 33 documented instances of hepatic cerebrospinal pseudocysts, with 15 cases specifically identified as intra-axial. The summary table [Table/Fig-7] presents a compilation of previous publications detailing reported cases of intra-axial hepatic cerebrospinal pseudocysts in individuals with VP shunts [3,5,10-17].

An intra-axial hepatic pseudocyst is formed when the VP shunt tube pierces the capsule, liver parenchyma, and gets lodged within the parenchyma with CSF getting collected surrounding the shunt tip, whereas an extra-axial CSF pseudocyst forms in the subcapsular space between the liver parenchyma and hepatic capsule when the shunt pierces only the liver capsule [10].

On a review of clinical features in 15 patients with hepatic CSF pseudocysts, it was found that more than 90% had abdominal symptoms and signs, such as abdominal pain, distension, and right upper quadrant tenderness, whereas less than 40% had symptoms of raised intracranial pressure [3]. In the present case, the patient had complaints of right upper quadrant pain and tenderness for a period of one month without features of raised intracranial pressure.

Imaging modalities for the evaluation of CSF pseudocysts include abdominal radiograph, ultrasound, Computed Tomography, and Magnetic Resonance Imaging. An abdominal radiograph may aid in the diagnosis by showing the course and location of the shunt and its distal tip [7], as seen in the topogram image of the present case. Ultrasound is a good screening tool as it is easy, inexpensive, and can be performed at the bedside without radiation exposure. Ultrasound demonstrates the size, location, and characterisation of the collection with the distal tip of the shunt within. Computed tomography scores over other modalities as it can demonstrate the entire course of the VP shunt with the exact location and size of the collection [7,10].

Due to the rarity of this complication, the management of hepatic CSF pseudocyst remains controversial, with the management options being patient-specific. In asymptomatic individuals with an incidental finding on imaging, the consensus is expectant care with routine monitoring using radiological imaging. Abdominal ultrasounds every one to two years can be performed in such patients to monitor the size of the pseudocyst and plan between conservative or surgical treatment options. In symptomatic individuals, initially, an imaging-guided diagnostic aspiration is done. If an infection is found, antibiotic therapy and shunt externalisation are performed. Later, repositioning of the shunt is done once the infection clears. The hepatic CSF pseudocysts, depending on the expertise available, can be treated by elective robotic/laparoscopic excision of the pseudocyst, debridement of

Authors	Year	Age	Sex	Cyst location	Aetiology of hydrocephalus	Treatment	Follow-up	Outcome
Current Case	2023	14	F	Intra-axial	Tumour	Laparoscopic drainage and cyst excision with shunt revision.	6 month	Complete resolution of the cyst.
Yousaf MN et al., [10]	2023	49	M	Intra-axial	Congenital	Robotic laparoscopic pseudocyst fenestration, partial hepatectomy, and a repositioning of VPS	4 weeks	Complete resolution of symptoms without any complications
Bettis T et al., [11]	2019	56	M	Intra-axial	Trauma	N/A	N/A	N/A
Dabdoub CB et al., [3]	2013	40	M	Intra-axial	Trauma	VPS removal with reinsertion and cyst drainage	36 week	No clinical recurrence of hepatic CSF pseudocyst.
Verma A et al., [12]	2012	35	M	Intra and extra-axial	Tumour	Pseudocyst aspiration and externalisation of shunt followed by reinsertion	3 week	Complete resolution of cyst on abdominal ultrasound.
Faraj W et al., [5]	2011	18	M	Intra-axial	Bacterial meningitis	Laparotomy and pseudocyst drainage followed by repositioning of shunt	N/A	Complete recovery.
Kolić Z et al., [13]	2010	30	F	Intra-axial	Subarachnoid Haemorrhage (SAH)	Ultrasound-guided aspiration and external ventricular drainage	4 weeks	Patient died due to sepsis 1 month after hepatic CSF pseudocyst drainage.
Aparici-Robles F and Molina-Fabrega R [14]	2008	22-50	F(4) M(2)	Intra-axial	Abscess, tumour, meningitis and SAH	Laparotomy with pseudocyst drainage and repositioning of shunt	-	There was a relapse in 2 of the 6 patients.
Chitkara N et al., [15]	2004	5	F	Intra-axial	Non communicating	Externalisation of shunt followed by reinsertion	3 month	Patient was entirely asymptomatic and cyst had completely resolved
Rana SR et al., [16]	1985	12	M	Intra-axial	Tumour	Repositioning of shunt	N/A	Complete resolution of cyst in follow-up CT scan
Latchaw Jr JP and Hahn JF [17]	1981	40	M	Intra-axial	Aqueductal stenosis	Drainage of hepatic pseudocyst and repositioning of the shunt.	N/A	N/A

[Table/Fig-7]: Summary table of reported cases of intrahepatic cerebrospinal pseudocyst including the current case [3,5,10-17].
{This table is modified from the table made by Yousaf MN et al., [10]}

the cystic wall, and/or repositioning the VP shunt catheter in the lower quadrant of the abdomen.

CONCLUSION(S)

Intra-axial hepatic CSF pseudocyst formation is a late and extremely rare complication of VP shunting. The clinical presentation, physical examination, and laboratory findings are often non specific and may confound the diagnosis. Imaging using ultrasound and CT is often the methods of choice to establish the diagnosis. The imaging features of the VP shunt tip within the cyst and the rim of liver parenchyma around the cyst should provoke the radiologist to consider a differential diagnosis of intra-axial hepatic CSF pseudocyst.

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