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Relapsing-Remitting Hepatic Pseudo-Cyst: a great simulator of malfunctioning ventriculoperitoneal shunt. Case report and proposal of a new classification.

Le pseudo-kyste hépatique récidivant: simulateur de dysfonctionnement de dérivation ventriculo-péritonéale. Rapport de cas et proposition d'une nouvelle classification.

C-H. Mallereau[1], M. Ganau[1], J Todeschi[1], P-F. Addeo[2], S. Moliere[3], S. Chibbaro[1]

1: Neurosurgery Department, Strasbourg University Hospital, Strasbourg, France.
2: General and Hepato-Gastric Surgery Department, Strasbourg University Hospital, Strasbourg, France.
3:*Radiology Department, Strasbourg University Hospital, Strasbourg, France.*

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Abbreviations:

ACoA : Anterior Communicating Artery
A&E : Accident and Emergency
CSF : Cerebrospinal Fluid
CT Scan : Computerized Tomography Scan
EVD : External Ventricular Derivation
MC&S: Microscopy, culture & sensitivity
NSAID: Non-Steroidal Anti-Inflammatory Drugs
SAH : Subarachnoid Hemorrhage
VAS : Ventriculoatrial Shunt
VPS : Ventriculoperitoneal Shunt

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Abstract

Background

Ventriculoperitoneal shunt is the most common treatment to manage hydrocephalus; It is unfortunately burdened by up to 25% of complications. The peritoneal approach may expose patients to many complications, however the formation of a liver pseudocyst is a rare occurrence, and its mechanisms are still largely unknown.

Case report

We report the case of a 69-year-old woman with ventriculoperitoneal shunt, inserted for the management of post aneurysmal subarachnoid hemorrhage hydrocephalus, presenting to the Accident and Emergency for acute cholecystitis. Besides confirming the diagnosis, an ultrasound investigation revealed the presence of a hepatic cyst. Conservative treatment with antibiotics and non-steroidal anti-inflammatory drugs was performed with favorable outcome and resorption of the cyst. Interestingly the patient kept on presenting several similar episodes managed well by non-steroidal anti-inflammatory drugs alone, each of them associated with transient symptoms and signs of ventriculoperitoneal shunt malfunction. Computerized Tomography brain and lumbar puncture were normal, whereas CT abdomen showed the ventriculoperitoneal shunt distal catheter passing through the hepatic cyst. Given the

ventriculoperitoneal shunt malfunction, in the context of an infective/inflammatory process a conversion of the ventriculoperitoneal shunt into a ventriculo-atrial shunt was carried out with successful clinical outcome.

Conclusion

Based on current literature we propose a clinical and radiological classification of such pseudocysts related to ventriculoperitoneal shunt. Clinical presentation, diagnostic findings and management options are proposed for each type: purely infective, spurious (infective/inflammatory) and purely inflammatory. In the absence of system infection, a simple replacement of the distal catheter seems to be the best solution.

Keywords: Hydrocephalus, Ventriculoperitoneal shunt, Hepatic pseudocyst, Abdominal surgery.

Résumé

Contexte

La dérivation ventriculo-péritonéale est un traitement fréquent de l'hydrocéphalie mais responsable de complications allant jusqu'à 25%. L'approche péritonéale peut exposer les patients à des complications particulières, mais la formation d'un pseudo-kyste hépatique est rare et son mécanisme méconnu.

Rapport de cas

Nous rapportons le cas d'une femme de 69 ans porteuse d'une dérivation ventriculopéritonéale, mise en place pour une hydrocéphalie post-hémorragie sous-arachnoïdienne, se présentant aux urgences pour une cholécystite aiguë. Une échographie a confirmé la cholécystite aigue et a révélé la présence d'un kyste hépatique. Un traitement médical par antibiotiques et d'anti-inflammatoires non stéroïdiens a permis un résultat favorable et une résorption du kyste. La patiente a toutefois présenté plusieurs épisodes similaires bien gérés par les anti-inflammatoires non stéroïdiens seuls, chacun d'eux étant associé à des symptômes transitoires de dysfonctionnement de dérivation ventriculo-péritonéale. Le scanner cérébral et la ponction lombaire étaient normaux, tandis que le scanner de l'abdomen a montré que le cathéter distal de la dérivation passait par le kyste

hépatique. Étant donné le dysfonctionnement du système évoluant dans le contexte de processus infectieux/inflammatoire, une conversion de la dérivation ventriculopéritonéale en ventriculo-atriale a été réalisée avec succès.

Conclusion

A partir de la littérature, nous proposons une classification clinico-radiologique de ces pseudo-kystes. La présentation clinique, les résultats du diagnostic et les options de gestion sont proposés pour chaque type : purement infectieux, inflammatoire survenant dans un contexte infectieux et purement inflammatoire. En l'absence d'infection du système de dérivation, un simple remplacement du cathéter distal semble être la meilleure solution.

Mots clés : Hydrocéphalie, dérivation ventriculopéritonéale, pseudokyste hépatique, chirurgie abdominale.

1. INTRODUCTION

Insertion of a ventriculo-peritoneal shunt (VPS) is the most common surgical option for the long-term management of hydrocephalus and represents one of the most frequent interventions in neurosurgery. Overall, VPS failure is estimated between 11-25% in the first year following the operation in an adult population [1–4]. The most common causes for shunt failure are obstruction/VPS dysfunction and infection [4, 5]. During the surgical steps of the peritoneal approach and tunneling of the catheter, many complications may be encountered, including: a) distal catheter malposition/ectopy/migration resulting in extra-peritoneal (subcutaneous/intramuscular) collection of cerebrospinal fluid (CSF), b) intra-abdominal formation of growing CSF pseudocyst and, c) peritonitis, d) intestinal obstruction, e), bladder and bowel perforation [6–8]. Less commonly, any iatrogenic or inflammatory lesion of a solid visceral organ, such as the liver or the spleen, may cause encasement of the distal catheter paving the way for its long-term failure[9–31]; this rare event can manifest with the formation of a proper pseudocyst [32], whose pathophysiological mechanisms are still being discussed [33]. We describe a very rare case of hepatic pseudocyst presenting by abrupt abdominal symptoms hiding an underlying chronic VPS dysfunction, providing also a new classification proposal based on current literature.

2. CASE REPORT

2.1.History

A 69-year-old lady with a past medical history consisting essentially of a post-hemorrhagic hydrocephalus due to ACoA aneurysmal SAH which had been managed by VPS in July 2015, in another institution abroad during annual holiday, was referred to our A&E for persistent abdominal pain. She presented with fever and raised inflammatory markers leading to the diagnosis of lithiasic cholecystitis, confirmed by hepatic ultrasound which showed also an incidental cyst involving the 6^{th} hepatic segment. The patient was successfully managed by antibiotics and NSAID; of note, the abdominal ultrasound performed before discharge revealed a resolution of the cholecystitis as well as the involution of the hepatic cyst. Two months later the patient presented once again the same abdominal symptoms without fever but a recurrent, biochemistry confirmed inflammatory syndrome. The abdominal CT scan realized showed the recurrence of the hepatic cystic lesion, demonstrating also that the distal catheter of the VPS was passing within it (Figure 1). A course of NSAID was immediately started leading to a progressive clinical improvement; as such, the patient preferred not to proceed with further surgical treatment despite the general surgeon's advice to consider a cholecystectomy as a more definitive treatment.

In light of the CT abdomen findings, the patient was also referred to the neurosurgical outpatient clinic, and a more careful clinical history taken; her relatives reported that the patient had been suffering from episodic gait and urinary sphincter disturbances, dating since several months, with more recent onset of memory impairment.

A brain CT scan did not show ventricular dilatation. After discussion with our hepatic surgery team, an ultrasound guided cyst biopsy/puncture was scheduled but could not be performed due to the disappearance of the lesion after just 3 weeks from the end of the NSAID treatment (Figure 2 and 3). A lumbar puncture did not show any abnormal finding in terms of CSF pressure or laboratory analysis, hence no VPS tapping was deemed appropriate.

2.2.Treatment

The case underwent collegial discussion in our multidisciplinary team meeting; a consensus was reached among the neurosurgical, general surgery and infectious disease teams regarding the hypothesis of a VPS dysfunction, prompting VPS revision with conversion to a ventriculo-atrial shunt (VAS) considering the spurious (inflammatory/infectious) context. The patient underwent elective surgery few weeks later; an ultrasound performed the eve of the procedure showed initial recurrence of the hepatic cyst, with the patient reporting once again a progressive appearance of gait and sphincter disturbances. The conversion of the VPS into a

VAS was uneventful and the CSF sample sent to laboratory for MC&S analysis returned entirely normal. Intraoperatively, the distal catheter was found partially blocked by fibrin thrombi and sent for culture that was negative for bacterial infection at 5 and 10 day checks. The postoperative course was uneventful with an immediate and remarkable improvement of the walking and sphincter problems, allowing a safe discharge home on the 4th postoperative day after a favorable CT scan (see also figure 4).

2.3.Follow up

A clinical and radiological follow up at 3, 6 and 12 months was unremarkable without any cyst recurrence and the patient reporting a generalized wellbeing.

3. DISCUSSION

3.1. Generalities

We report a case of a secondary hepatic pseudocyst, resulting from a pauci-symptomatic VPS malfunctioning, successfully managed by conversion of the VPS into a VAS. Pseudocyst formation is certainly a relatively rare occurrence, often resulting in shunt blockage or infection, as well as other abdominal problems.

3.2.Pseudocyst types

This complication comes across as a very heterogeneous one, both in terms of its presentation and anatomical location. Hepatic pseudocyst can be classified as: a) intra-axial, when the tip of the shunt can be found/lodged into the liver and causes the intrahepatic cyst formation [21]; and b) extra-axial, when the tip of the catheter may enter the Glisson capsule but not the hepatic parenchyma, resulting in the formation of a subcapsular pseudocyst [13], as happened in our case. Given the different management needed in relation to the cyst type, we made a structured classification depending on their pathological context (see also table 1).

The lesions containing obvious pus (found on ultrasound or CT guided puncture) and clinically characterized by symptoms and signs of infection can be classified as purely infective (abscess), those accounted for 39% cases [9,12,14–16,18–20,30]. Management

consists mostly of externalization of the system, associated with cyst puncture to target antibiotic therapy, followed by delayed reinternalization [9,12,15,18–20].

On the other hand, true pseudocysts could be further separated into two groups: Spurious pseudocyst, non-abscessed cystic inflammatory lesions whose contents are not purulent but occurring on a subacute infection (peritonitis, meningitis) representing 17 % of cases [22,25,27,29]. In this lesional setting, we suggest, in agreement with current literature, that when the CSF is infected, an externalization of the system is recommended with re-internalization after sterilization of CSF whereas when there is no meningitis, a conversion from VPS to VAS appears sufficient to avoid subsequent/persistent inflammatory adhesions and/or favoring chronic infection [27].

The purely inflammatory pseudocysts, without clinical or laboratory evidence of infectious signs, constituting the 39% of cases in which the simple replacement of the distal catheter is suggested [10,11,13,17,23,24,26,28,31].

It is interesting to note that in the literature, not all patients with liver abscesses or pseudocysts evolving in an infectious context had an EVD while waiting for the complete resolution of their infection, whereas one of the patients with a spurious inflammatory/infectious pattern died while undergoing antibiotic treatment on EVD[25]. This

demonstrates that this condition requires a strict monitoring as the pseudocyst could behave as a great simulator thus its potential seriousness could be underestimated. We would therefore suggest that all infected and spurious pseudocyst should be treated as systemic infection prompting externalization of the shunt, whereas purely inflammatory ones should be considered as loco-regional complications requiring only the revision of the distal catheter; in case of clinical suspicion of infection the puncture/biopsy/culture of the cyst content could be considered (see also Table 1). The observation that the large majority of these cysts respond well to such treatment reinforces the pathophysiological hypothesis of a local inflammatory and/or mechanical injury.

3.3. Physiopathology

The pathological mechanisms underlying the formation of an hepatic pseudocyst are unclear, although it could be speculated that they are related to a chronic inflammatory process seemingly to the abdominal pseudocysts formation [9,21,34–36]. Chronic or sequential inflammation of the peritoneum or multiple surgeries could play a role in this process by anchoring the distal end of the catheter to a visceral structure, causing mechanical irritation and necrosis leading in the long term to its perforation[27,30].

Multiple shunt revision could also be a predisposing factor, although in the literature, shunt revision has been found related only in 45% of cases [29].

4. CONCLUSION

Hepatic pseudocysts are very rare complications of a VPS. In patients previously treated with VPS for long-term management of hydrocephalus, an hepatic pseudocyst can be found in the context of abdominal symptoms associated with fever with or without clinical signs of VPS malfunctioning. As such, specific/dedicated imaging is warranted to reach a definitive diagnosis. Although it is not possible to draw clear management recommendations, the use of the proposed classification can guide clinicians in understanding the pathophysiology and possibly select the most appropriate treatment options. The management of the pseudocyst, especially the purely inflammatory ones, should be tailored to the patients' presentation/condition and clinicians' choice, preference and experience. On the other hand, whenever an infective condition is confirmed, a more aggressive treatment with insertion of EVD, removal of the entire VPS, evacuation of the abscess and prolonged antibiotic therapy before considering shunt reinsertion is recommended.

Declarations

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Conflict of interest : none.

Authors' contributions :

This is to certify that all authors have participated in the present study including its conception, writing and critical revision.

Legends

Figure 1: Axial (A), Coronal (B) and 3D reconstruction (C) of enhanced Abdominal CT scan showing a subcapsular cystic lesion with a tick and slightly enhancing wall in the posterior aspect of the VI hepatic segment next to the Morrison's space in contact with the VPS distal catheter suggestive of an inflammatory process.

Figure 2: Check Hepatic MRI after 3 weeks of antibiotic and NSAID therapy showing regression of the lesion not allowing the scheduled puncture/biopsy.

Figure 3: Brain CT scan showing no evidence of hydrocephalus/ventricles dilatation despite a symptomatic clinical syndrome.

Figure 4: 48 hours postoperative check Brain CT scan showing no hemorrhagic complications, the correct VPS material position although the ventricular cavity size is unchanged

Table 1. Cysts features and their suggested management.

Table 1 Legend: CSF: Cerebro Spinal Fluid; VAS :Ventriculo-Atrial Shunt;

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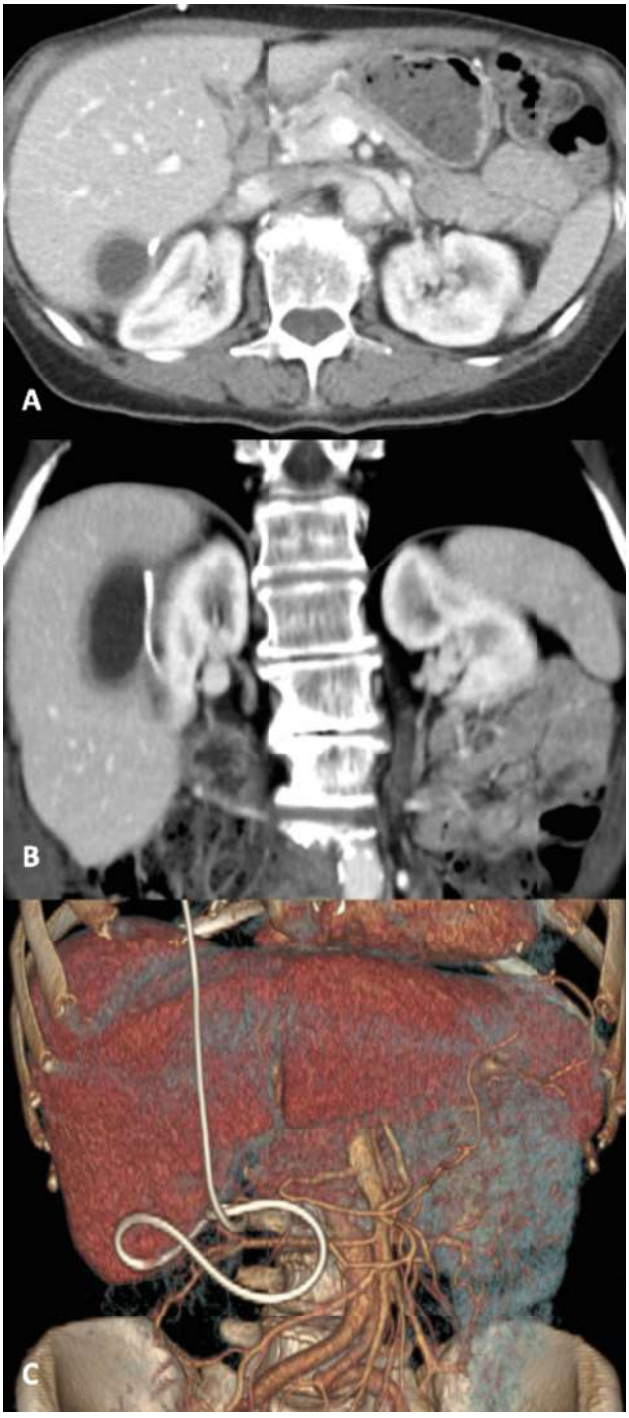
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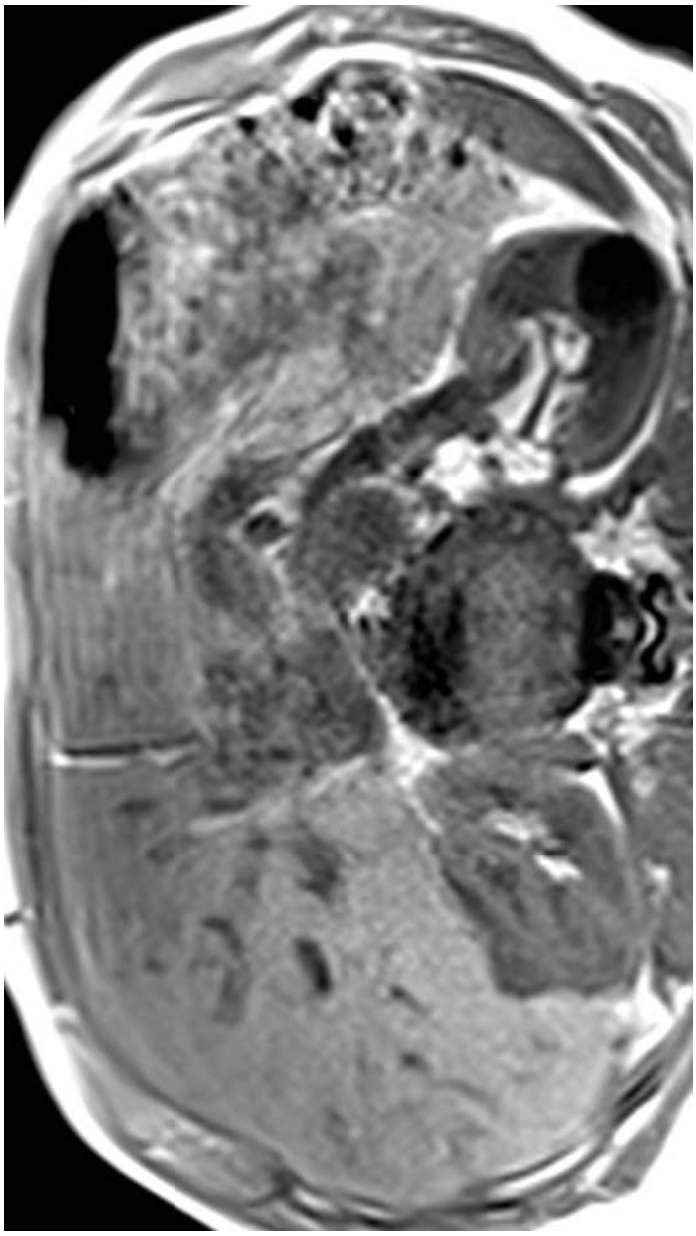

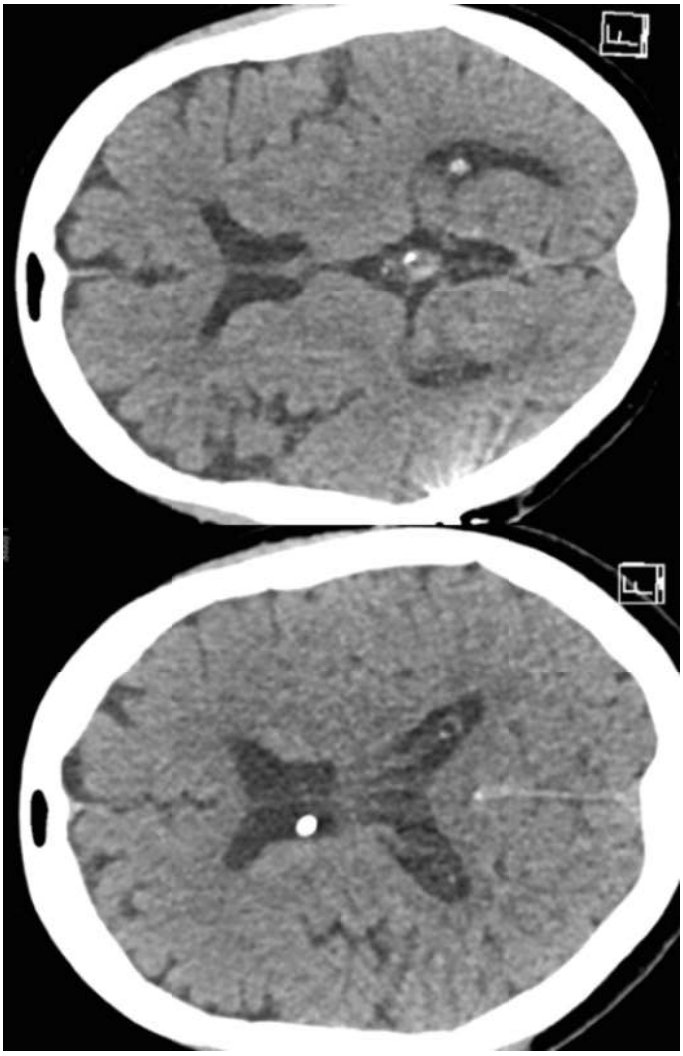

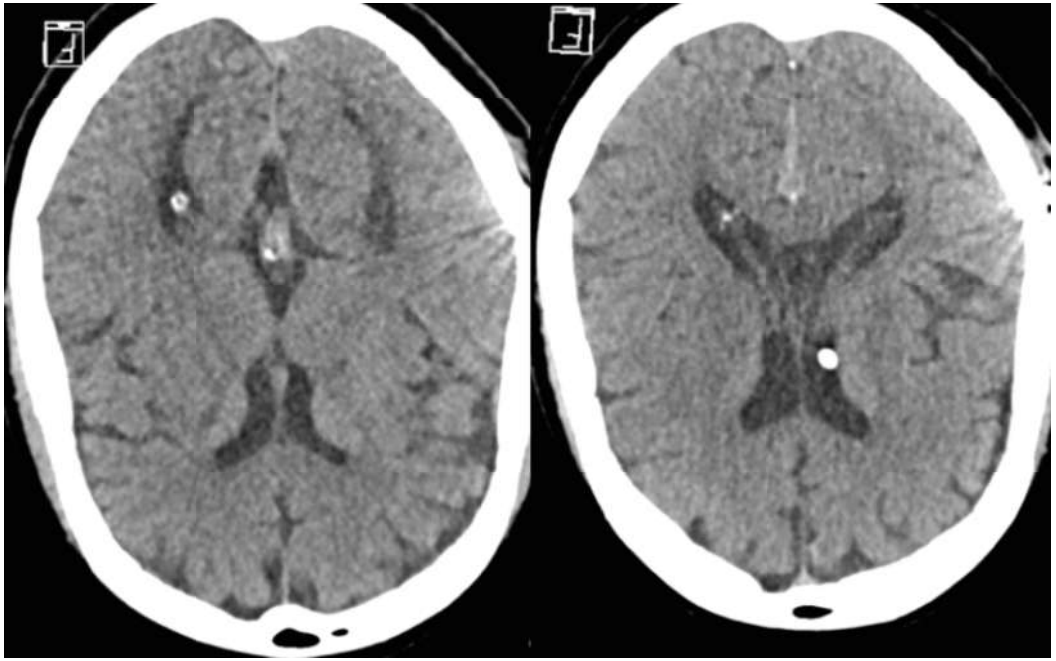

	Infected pseudocyst (abscess)	Spurious Pseudocyst (infectious/inflammatory)		True inflammatory Pseudocyst
		CSF negative	CSF positive	
Features	Frank pus found on puncture.	Inflammatory signs and positive culture in CSF or blood cultures without frank pus on puncture.		No clinical or laboratory evidence of infection
Intra-axial pseudocyst (intra-hepatic)	Shunt and cyst lodged in the hepatic parenchyma[21]			
Extra-axial pseudocyst (intracapsular)	Shunt and cyst lodged in the Glisson capsule in the peri-hepatic space (extra-hepatic) [13]			
Suggested management following rigorous analysis of available literature	Externalization of the system and cyst puncture followed by delayed VPS reinternalization [9,12,15,18–20]	Conversion into a VAS [27]	Externalization of the system and cyst puncture followed by delayed VPS reinternalization [22,25]	Replacement of the distal catheter in the opposite side [10,11,13,17,23,24,26,28]