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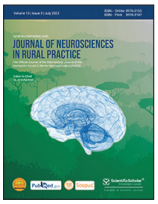

Case Series

Central dilemma in CSF pseudocyst – A case series and review of literature

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ABSTRACT

Cerebrospinal fluid (CSF) pseudocyst abdomen is a rare but well-described complication following ventriculoperitoneal (VP) shunt. This entity does exist since 1954. This is seen more commonly among pediatric population and cause of CSF pseudocyst is still debated, many theories been postulated in the literature and so are its management. We present our experience with small case series and idea is to provide an alternate management strategy for shunt-independent cases. We did retrospective study of three cases, diagnosed on the basis of clinical profile and imaging. Subclinical infection was ruled out and patients with abdominal complaints predominantly and no ventriculomegaly on Noncontrast computed tomography head were subjected to "shunt-tie" at infraclavicular region. Out of three cases, two had abdominal complaints with no features of raised ICT and no ventriculomegaly. On tying the shunt catheter infraclavicular level for 48–72 h, they did not developed raised ICT/ventriculomegaly. Cyst was drained by percutaneous ultrasound-guided PIGTAIL. Shunt assembly was removed. One patient (shunt dependent) underwent exploratory laparotomy and repositioning of the catheter but experienced shunt malfunction, ultimately VP shunt was converted to ventriculopleural shunt. On follow-ups, there is no residual cyst or recurrence of symptoms. To conclude, evaluation of shunt dependency/non-dependency is of utmost importance. For shunt-independent cases, percutaneous ultrasound-guided PIGTAIL drainage is safe, minimally invasive, and effective procedure and we may avoid many potential complications.

Keywords: Ventriculoperitoneal shunt, Pseudocyst abdomen, Management dilemma, Shunt dependent

INTRODUCTION

Ventriculoperitoneal (VP) shunt surgery is one of the most common procedures performed in neurosurgery. Cerebrospinal fluid (CSF) pseudocyst abdomen following VP shunt is an uncommon complication with incidence ranging from 0.33% to 6.6%.[1-3] This entity was first reported by Harsh in 19541–3. This complication is mostly seen among children and is extremely rare in adults.

There have been varied management strategies of an abdominal pseudocyst. Most of the strategies have focused on laparotomy and repositioning of the abdominal end of the shunt to another abdominal quadrant without considering if the patient is shunt dependent or independent. Our study aims to provide an alternate management strategy by tying the abdominal catheter of the shunt and draining pseudocyst with ultrasound-guided PIGTAIL. This strategy is minimally invasive, safe, and may avoid laparotomy-related additional complications in most cases.

CASE-WISE DETAILS

Case wise details in Table 1.

DISCUSSION

Common abdominal complications following VP shunt surgery are peritonitis, ascites, bowel or abdominal wall perforation, and inguinal hernias.[1,4,5] Pseudocyst is a collection of CSF around terminal catheter [Figure 1], which grows slowly over a long time. Some patients have reported this complication after 3–4 weeks but a few may present after many years (3 weeks–10 years).[6]

Etiological aspects

There is no well-established etiology for the CSF pseudocyst in the literature. Long-standing subclinical abdominal infection,[7,8] high-protein contents in CSF, recurrent abdominal surgeries, increased abdominal adhesions, and silicone allergy[9] are some of the contributing factors for decreased CSF

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Table 1: Case-wise details.

S. No.	Age/gender	Presentations	Duration (shunt surgery)	CSF analysis/inflammatory markers	Signs of increased ICP post-ligation of catheter (shunt dependency/independence)	Definitive treatment	Complications	Follow-ups	Recurrence
1.	31 years/male	Right hypochondrium and epigastric pain with abdominal distension	15 years back	WNL	No (shunt independent)	Ultrasound-guided percutaneous pigtail drainage	Nil	6 months, 12 months, and 24 months	No
2.	34 years/male	Epigastric discomfort with abdominal distension	Few years back	WNL	Yes, after 2 days (shunt dependent)	Exploratory laparotomy with excision of cyst with repositioning of catheter. He developed raised ICP features. Finally, underwent ventriculopleural shunt placement	Pleural effusion (after ventriculopleural shunt) which was managed conservatively	6 months and 12 months	No
3.	10 months/male	Abdominal distension	Few months Back	WNL	No (shunt independent)	Ultrasound-guided percutaneous pigtail drainage	Nil	6 months, 12 months, and 24 months	No

WNL: Within normal limits

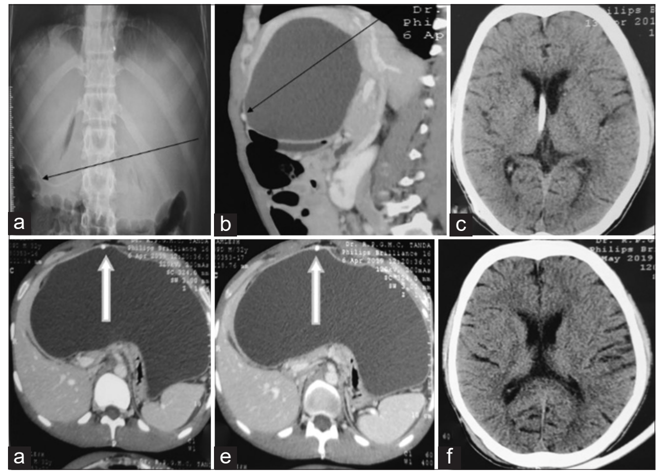

Figure 1: An X-ray showing the abdominal end of the shunt (black arrow) in the location of pseudocyst (a). Noncontrast computed tomography (NCCT) abdomen showing large pseudocyst abdomen with abdominal catheter lying along its anterior wall, as shown by black and bold white arrows (b and d). NCCT head showing the ventricular end of the shunt (c). NCCT head after removal of the shunt with no hydrocephalous (e).

absorption, leading to CSF collection in the form of pseudocyst formation. A subclinical infection has been documented in 17–80% of cases.[10] In addition, few pathogens have also been found quite commonly associated with pseudocyst which include *Staphylococcus epidermis*, *Staphylococcus aureus*, and *Streptococcus*.[10,11] At times, the subclinical infection caused by shunt may not be diagnosed by single CSF culture and infection may remain latent.[5,11] In our cases, cultures were all sterile presuming the role of subclinical infection.

Clinical manifestations

We noticed abdominal distension on presentation, which was insidious in onset and gradually progressive in all three cases [Figure 1] and case-wise details are mentioned in [Table 1]. Associated features of raised ICP such as headache, nausea, and vomiting may be seen especially in shunt-dependent cases (as in one of our cases) whereas abdominal distension with or without pain may be the only presenting symptom, especially in adult patients. Other less common features may be fever, anorexia, constipation, tenderness, palpable mass, and subphrenic abscess.[10]

Management dilemma

Management of CSF pseudocyst should be tailored to individual patient as there is no standard/uniform management strategy defined for pseudocyst abdomen and is always a management dilemma [Figure 2]. The options discussed in the literature [Table 2] include open procedures such as laparotomy and cyst drainage/cyst wall

Table 2: Comparative evaluation and management strategies by various authors in the literature.

Authors	Year	Journal and number of cases (*n*)	Management options	Role of shunt dependency	CSF/WBC/ESR levels	Complications
Hamid *et al.*	2017	Asian journal of neurosurgeon (*n*=4)	Cyst excision with repositioning of catheter	Not analyzed	Not analyzed	Not mentioned
Yuh and Vassilyadi	2012	Surgical neurological international (*n*=1)	Cyst drainage with ventriculopleural shunt	Not analyzed	-	Pleural effusion
Kashyap *et al.*	2017	Surgical neurological international (*n*=4)	USG-guided cyst drainage and shunt exteriorization and internalization	Not analyzed	Yes	Not mentioned
			USG-guided aspiration and VA shunt	-	Yes	Not mentioned
			USG-guided drainage of cyst and ETV		Yes	ETV failure
Masoudi	2017	African Journal of Pediatric Surgery (*n*=1)	Laparotomy and cyst marsupialization Abdominal catheter repositioning followed by VA shunt	Not analyzed	Yes	Recollection/ recurrent pseudocyst abdomen
Anwar *et al.*	2017	BMJ (*n*=1)	Shunt externalization and placement of new shunt	Not analyzed	No	Not mentioned
Popa *et al.*	2009	J Med Life (*n*=2)	Evacuation of cyst with resection of cyst wall with drainage of peritoneal cavity. EVD followed by VA shunt	Not analyzed	CSF analyzed	Not mentioned
Wang *et al.*	2021	BMC, Surg (*n*=1)	Laparoscopic evacuation with excision of while cyst. No shunt revision (distal shunt catheter discarded)	Not analyzed	Not analyzed	Not mentioned
Mobley *et al.*	2005	Pediatric neurosurgery (*n*=64)	Pre-operative aspiration of CSF from shunt and cyst followed by +/- antibiotics and VP shunt or VA or VPL	Not analyzed	CSF and TLC analyzed preoperatively	Not mentioned
Tomiyama *et al.*	2014	Hindawi Publishing Corporation Surgery Research and Practice (*n*=1)	Drainage of infected cyst with EVD using same ventricular end followed by antibiotics and VP shunt	Not analyzed	CSF analyzed	Not mentioned
Koide *et al.*	2019	Journal of Medical Case Reports (BMC) (*n*=1)	Cut abdominal end at chest , removed distal end and used proximal end as EVD followed by percutaneous drainage and VP shunt in new quadrant	Not analyzed	CRP/TLC analyzed	Not mentioned
Roitberg *et al.*	1998	Pediatric neurosurgery (*n*=27)	Removal of shunt, EVD followed by antibiotics. Cyst aspirated and new shunt in different quadrant	Not analyzed	CSF workup only	Revision surgery (*n*=4) Recurrence of pseudocyst (*n*=2)
Salomão and Leibinger	1999	Pediatric neurosurgery (*n*=18)	Removal of distal catheter and exteriorization (*n*=04) EVD (*n*=11), catheter repositioning (*n*=2), laparoscopic excision of cyst (*n*=1) followed by VA in all	Not analyzed	CSF workup only	Recurrence (*n*=3)
Gaskill and Marlin	1989	Pediatric neurosciences (*n*=12)	Shunt removal followed by VA/VPL or EVD/antibiotics. Cyst resorbs spontaneously	Not analyzed	CSF workup only	No recurrence

ETV: Endoscopic third ventriculostomy, EVD: External ventricular drainage, VPL: Ventriculopleural, VA: Ventricular atrial, VP: Ventriculoperitoneal, CSF: Cerebrospinal fluid

excision or laparoscopic cyst drainage with repositioning of the abdominal catheter of the shunt.[5,11-15] The laparoscopic method is considered a preferred method for pseudocyst abdomen management by many authors. Other options such as exteriorization of abdominal end with or without concomitant antibiotics and the alternate site or contralateral side for shunt catheter placement are suggested by many. Whereas few authors have advocated ventriculoatrial (VA), ventriculopleural (VPL), or endoscopic third ventriculostomy, especially when peritoneal cavity is not a suitable environment or infection free.[11,16] A comparative evaluation of management is detailed in Table 2.[5,11-15,17-21]

Open procedures such as laparotomy and excision of pseudocyst abdomen have been associated with multiple adhesion formation, obstruction, wound infections, etc. Such procedures are prone to recurrence of pseudocyst. Whereas, VPL shunt placement is a good alternative but it carries the risk of pleural effusion [Table 2], as in our Case 2. Most of the management options do not address the importance of clinical or subclinical infection, which may be a contributing factor. However, it is critically important that the presence or absence of subclinical or on-going infection as an infected assembly may continue to be a nidus.[3,11] Laparoscopic excision of the cyst and repositioning of the catheter are a less morbid procedure but we have observed in shunt-independent/non-dependent cases, even such procedures can be avoided.

Most of the studies have not taken into account of the role of CSF workup or other inflammatory markers (TLC, ESR, and CRP) before proceeding any intervention. These levels are necessary as they will guide whether to exteriorize the shunt or not. Similarly, antibiotics need to be started as per culture and sensitivity.[22]

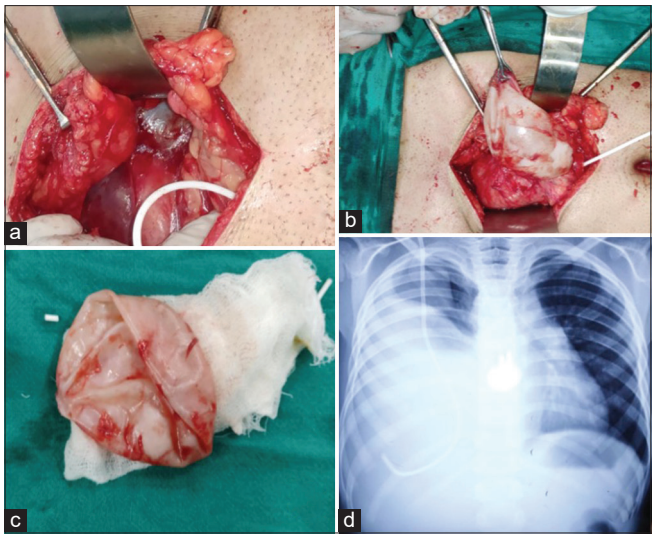

Figure 2: Exploratory laparotomy with collapsed pseudocyst abdomen and shunt tip (a-c). Development of pleural effusion following ventriculopleural shunt (d).

Role of shunt dependency – A key step

To the best of our knowledge, none of the studies has given any emphasis on shunt dependency. The "shunt dependency" has to be ruled out right at the beginning, especially when there are no signs of raised ICP and only complaint is abdominal distension. We can rule out "shunt dependency" by a simple and safe technique. One needs to tie the abdominal catheter of the shunt just below the clavicle by making a small skin incision (1 cm) over the shunt catheter and further dissection to locate shunt catheter. One needs to avoid any accidental cut in underlying catheter. The shunt is tied with nylon 2–0 suture [Figure 3a-c]. The patient should be observed for 2–3 days for any feature of raised ICP. Repeat Noncontrast computed tomography head should be done after a couple of days (after 48–72 h). If there are no features suggestive of raised ICP clinically as well as radiologically, then we consider him/her "shunt independent" and it makes management strategy simpler thereafter. We can simply remove the whole shunt assembly as an offending source to eliminate the possibility of latent or subclinical infection. CSF pseudocyst abdomen is further managed by a very simple, effective, and minimally invasive technique, that is, ultrasound-guided percutaneous drainage of the cyst (PIGTAIL drainage), as shown in algorithm in [Figure 4]. Furthermore, this technique may avoid many expected complications such as pleural effusion following VPL shunt, chances of infection following exteriorization of shunt end, and laparotomy-related complications. Gaskill and Marlin

Figure 3: Child showing collapsed pseudocyst lump size after PIGTAIL drainage (on the right lower abdomen shown by black arrow) and site of tying shunt catheter infraclavicular, right side – shown by black arrow (a). Noncontrast computed tomography (NCCT) head showing no ventriculomegaly after tying shunt (b). Before draining, pseudocyst abdomen with shunt tips lying anteriorly and inferiorly (c).

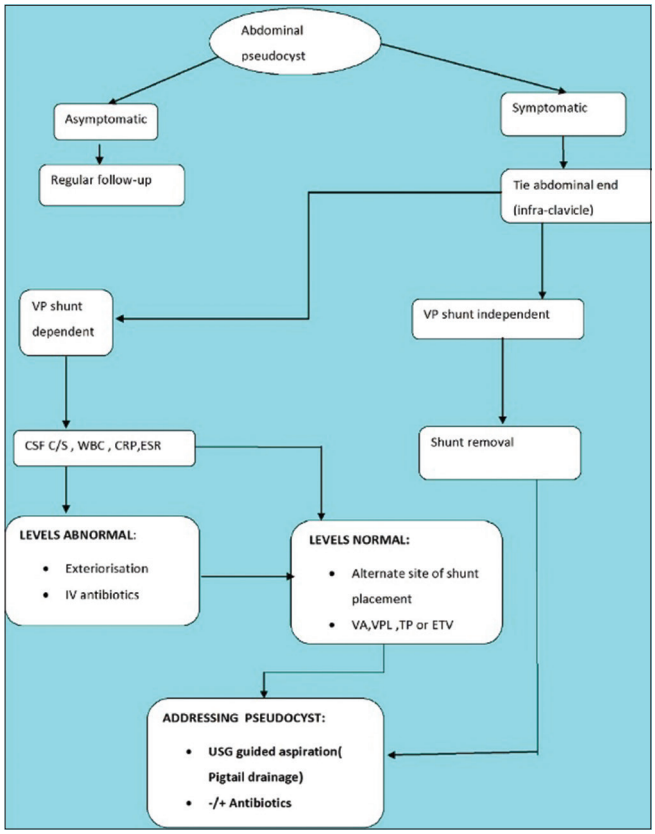

Figure 4: Proposed algorithm showing management strategies for shunt dependent and shunt non-dependent abdominal CSF pseudocyst.

documented in their series that it is not mandatory to remove the walls of pseudocyst because pseudocysts solve spontaneously, once the catheter is taken off.[23] In our cases, it has proven a successful modality and no recurrence is seen after 3-year follow-up.

In the case of shunt-dependent patients, management is always challenging as there are possibilities of failure or recurrence of the pseudocyst. Again analysis of inflammatory markers (TLC, CRP, and CSF) beforehand is of utmost importance. In case of recurrence, we need to choose other options such as VPL or VA shunt placements. Some authors also suggest placement of the distal catheter in the gallbladder. For infected cases, we need to exteriorize the distal end or convert the proximal end of the catheter into EVD and start broad-spectrum/culture-based IV antibiotics for 2–3 weeks or till two cultures are sterile [Figure 4].

CONCLUSION

Abdominal CSF pseudocyst is a rare complication following the VP shunt procedure, especially in adults. Management needs to be tailored according to various parameters. CSF and inflammatory markers are of great help in deciding treatment plan. To ascertain beforehand, shunt dependency/non-dependency is a key step to avoid potential complications. For shunt-independent pseudocyst, ultrasound-guided percutaneous drainage of a cyst (pigtail drainage) is a safe and effective method of treating pseudocyst abdomen.

In our study, despite limited number of cases, we had favorable outcome in such cases. In our opinion, more number of cases need to be evaluated to validate this management strategy (pigtail drainage) for shunt-independent cases to avoid laparotomy and alternate sites (VA and VPL) shunt placement-related complications.

Statement of ethics

Written informed consent was obtained from the patients for publication of this case report and any accompanying images. Ethical approval was not required for this study in accordance with national guidelines.

Data availability statement

All data generated or analyzed during this study are included in this article. Further enquires can be directed to the corresponding author.

Declaration of patient consent

Patient's consent not required as there are no patients in this study.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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