SNI

SURGICAL NEUROLOGY INTERNATIONAL

OPEN ACCESS

For entire Editorial Board visit : http://www.surgicalneurologyint.com

Editor: **James I. Ausman,** MD, PhD University of California, Los Angeles, CA, USA

SNI: General Neurosurgery

Case Report

Abdominal pseudocyst as a complication of ventriculoperitoneal shunt placement: Review of the literature and a proposed algorithm for treatment using 4 illustrative cases

Samir Kashyap, Hammad Ghanchi, Tanya Minasian, Fanglong Dong[1], Dan Miulli

Department of Neurosurgery, Riverside University Health System - Medical Center, Cactus Avenue, Moreno Valley, CA, [1]Department of Biomedical Science, Graduate College of Biomedical Sciences, Western University of Health Sciences, Pomona, California, USA

E-mail: *Samir Kashyap - skashyap41@gmail.com; Hammad Ghanchi - hammadghanchi@gmail.com; Tanya Minasian - tminasian@gmail.com; Fanglong Dong - fdong@westernu.edu; Dan Miulli - miullid@armc.sbcounty.gov
*Corresponding author

Received: 28 August 16 Accepted: 27 October 16 Published: 10 May 17

Abstract

Background: Ventriculoperitoneal (VP) shunt placement is one of the most commonly performed procedures in neurosurgery. One rare complication is the formation of an abdominal pseudocyst, which can cause shunt malfunction.

Case Descriptions: We present four unique cases of abdominal pseudocyst formation. Our first patient initially presented with a right upper quadrant pseudocyst. Shunt was externalized and the distal end was revised with placement of catheter on the opposite side. He developed another pseudocyst within 5 months of shunt revision and developed another shunt failure. Our second patient had a history of shunt revisions and a known pseudocyst, presented with small bowel obstruction, and underwent laparotomy for the lysis of adhesions with improvement in his symptoms. After multiple readmissions for the same problem, it was thought that the pseudocyst was causing gastric outlet obstruction and his VP shunt was converted into a ventriculopleural shunt followed by percutaneous drainage of his pseudocyst. Our third patient developed hydrocephalus secondary to cryptococcal meningitis. He developed abdominal pain secondary to an abdominal pseudocyst, which was drained percutaneously with relief of symptoms. The fourth patient had a history of multiple shunt revisions and a previous percutaneous pseudocyst drainage that recurred with cellulitis and abscess secondary to hardware infection.

Conclusion: Abdominal pseudocysts are a rare but important complication of VP shunt placement. Treatment depends on etiology, patient presentation, and clinical manifestations. Techniques for revision include distal repositioning of peritoneal catheter, revision of catheter into pleural space or right atrium, or removal of the shunt completely.

Key Words: CSFoma, pseudocyst, ventriculoperitoneal shunt

Access this article online

Website: www.surgicalneurologyint.com

DOI: 10.4103/2152-7806.206007

Quick Response Code:

How to cite this article: Kashyap S, Ghanchi H, Minasian T, Dong F, Miulli D. Abdominal pseudocyst as a complication of ventriculoperitoneal shunt placement: Review of the literature and a proposed algorithm for treatment using 4 illustrative cases. Surg Neurol Int 2017;8:78.
http://surgicalneurologyint.com/Abdominal-pseudocyst-as-a-complication-of-ventriculoperitoneal-shunt-placement:-Review-of-the-literature-and-a-proposed-algorithm-for-treatment-using-4-illustrative-cases/

INTRODUCTION

Ventriculoperitoneal (VP) shunts are a staple procedure in the neurosurgical field. Its indications vary but the principle of shunting cerebrospinal fluid from the ventricles into the peritoneal space was first introduced in 1908.[3,4,11,14] Complications include obstruction in the proximal catheter, valve, distal catheter, hernia, peritonitis, cerebrospinal fluid (CSF) ascites, infection, valve failure, disconnection, or breakage of the catheter. Abdominal pseudocyst (APC) formation is a rare complication with reports in literature varying from 0.25% to 10%.[2,5,10,13,16] These pseudocysts are often found in the pediatric population, however, this is likely due to the increased incidence of hydrocephalus in children. Some are benign and cause no symptoms whereas some can result in significant abdominal pain and even malfunction of the shunt system.

Here, we present four unique presentations of abdominal pseudocyst formation as a complication of VP shunt placement and provide an algorithm for the management of this known, but rare, complication.

CASE REVIEW

Case 1

A 32-year-old male presented to our service with complaints of headache, nausea, vomiting, and a "funny feeling" is his left abdomen, which had been progressing for the past 3 weeks. Patient worked as a commercial truck driver and had been experiencing increasing symptoms when driving at higher altitudes, with some resolution of symptoms as he descended. Patient's past medical history was significant for premature birth with a VP shunt placed when he was approximately 2-month-old for unknown reasons. Patient reported that his first shunt revision occurred in 2009 where his shunt was revised to a right frontal VP shunt. Patient later required a revision at an outside hospital in 2015 because the shunt was complicated distally with a pseudocyst. At that time, the distal portion of the catheter was replaced. Pseudocyst cultures did not grow any organic species.

On previous admission to an outside hospital, patient presented with a 3-week history of abdominal pain, gradually worsening. No headaches, nausea, or vomiting were reported at that time. Computed tomography (CT) scan of the abdomen revealed a 13 cm pseudocyst along the liver margin [Figure 1], with the distal tip of the catheter within the cyst. At that time, patient's shunt was externalized, and the cyst was drained by interventional radiology. Cultures from the pseudocyst were negative. Patient was placed on cefepime empirically while in the hospital. On day 22, shunt was inserted with the assistance of general surgery, using laparoscopic guidance. A choice was made to internalize the shunt in the left upper quadrant. Of note, upon entrance into the abdominal cavity, the descending colon was found to be adherent to the left anterior abdominal wall. Lysis of adhesions was performed with blunt dissection. There was no evidence of bowel injury or significant bleeding. Catheter was passed into the peritoneum and the incision was closed in the usual manner.

On admission to our hospital 1 year later, patient's CT of the abdomen demonstrated a 10-cm fluid-filled cyst around the distal end of the catheter. On admission, erythrocyte sedimentation rate (ESR) was 39 and C-reactive protein (CRP) was 2.13. CT of the head demonstrated no evidence of hydrocephalus, however, there were no other images for comparison. A lumbar puncture was performed, which demonstrated an opening pressure greater than 40 cm H_2O. CSF studies were negative for infection. Next, the patient's shunt was tapped. Proximal (intracranial) catheter demonstrated significant resistance and was unable to be drained. Distal catheter flushed with ease and demonstrated a pressure of 13 cm H_2O, with a manometer held at EAM. CSF studies from this tap demonstrated gram positive rods on gram stain, however, cultures were negative. A decision was made to externalize the shunt (proximal catheter, valve, and distal catheter) and place an external ventricular drain. Patient was placed on vancomycin at the recommendation of infectious disease. Three negative sets of CSF were drawn, each 2 days apart, before returning to the operating room for shunt revision. He was taken to the operating room with the assistance of general surgery for removal of the orphan catheter in his abdomen with the placement of a new catheter.

Case 2

A 32-year-old Caucasian male presented with a history of cerebral palsy and other cognitive deficits at birth with a VP shunt placement as well as during childhood. His cognitive baseline was at the fourth grade level. The patient and his caregiver stated that he has had multiple

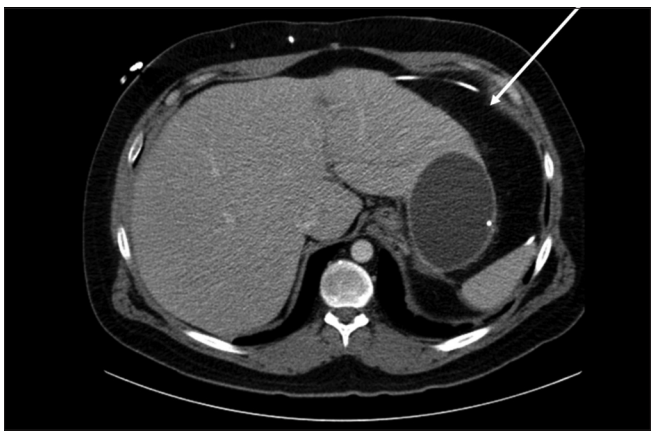

Figure 1: (Case 1) Computed tomography of the abdomen on the second admission with pseudocyst (arrow) in left upper quadrant

revisions in the past, which were performed at an outside hospital. In addition, he had a history of recurrent small bowel obstructions, for which he underwent a laparotomy for repair at an outside facility in 2007. He had two subsequent obstructions that resolved with bowel rest. He was initially admitted to our facility in October 2015 with abdominal pain, nausea, and vomiting, and was diagnosed with another small bowel obstruction. ESR on admission was 108; CRP was 14.30. CT of the abdomen at the time showed a pseudocyst in the abdomen measuring 15 × 14 × 10 cm [Figure 2] near the gastric outlet surrounding the peritoneal portion of the catheter. He was taken to the operating room by the general surgery team for lysis of adhesions. One month later, he presented to the emergency room with another small bowel obstruction, which was managed conservatively with resolution of his symptoms. Repeat CT abdomen showed a stable pseudocyst in the abdomen. He was discharged home, but returned to the ER again 1 month later with similar symptoms. Again, CT of the abdomen showed a stable pseudocyst near the gastric outlet. At this time, Neurosurgery was consulted because it was thought that the pseudocyst could be causing obstruction of the gastric outlet.

It was decided to convert his VP shunt into a ventriculopleural shunt. He underwent removal of the distal portion of the catheter where the peritoneal portion was removed and a new distal catheter was placed in the pleural cavity. He tolerated this procedure well without complication. It was then recommended that the patient have the abdominal pseudocyst aspirated by interventional radiology. Subsequent studies grew no organisms.

Case 3

A 40-year-old male with a history human immunodeficiency virus (HIV)/acquired immunodeficiency syndrome (AIDS) and hydrocephalus secondary to cryptococcal meningitis presented to the emergency room with diffuse abdominal pain and three episodes of nausea and vomiting. White blood cell count was elevated at 15.9 on admission, however, ESR and CRP were not obtained. CT head was performed on admission, which showed a narrowing of the lateral ventricles in comparison to previous CT. Neurosurgery and Infectious Disease were consulted for evaluation and a lumbar puncture was recommended. CSF was sent for gram stain and culture, which were negative. A CT of the abdomen was obtained which showed a large pseudocyst measuring 8.4 × 4 cm [Figure 3]. General surgery was then consulted for further management. It was their recommendation in combination with Neurosurgery that initial management should be percutaneous drainage by interventional radiology. The pseudocyst drainage was performed percutaneously, which resulted in near complete relief of patient's symptoms. Gram stain and culture from the fluid grew *Streptococcus Group D* that was pan-sensitive. On admission, he had been empirically started on Unasyn, of which he had completed a 5-day course prior to the procedure. He was discharged home on a 2-week course of Augmentin (875 mg BID).

Case 4

A 43-year-old female presented with a history of VP shunt placed at birth. She had multiple revisions throughout her lifetime. She presented to our service with abdominal pain, severe headaches, mild nausea, but no vomiting, and was found to have worsening hydrocephalus secondary to abdominal pseudocyst [Figure 4]. The cyst was determined to be infectious and was drained percutaneously; however, fluid from the pseudocyst and CSF grew *Streptococcus Group D*. Her shunt was externalized and eventually revised to the right side. One month later, she had a recurrent infection with the same organism. Shunt was externalized again and replaced after Infectious Disease consultation, and separate CSF cultures showed that the infection was cleared. She tolerated the procedure well and was discharged home in stable condition with alleviation of her symptoms.

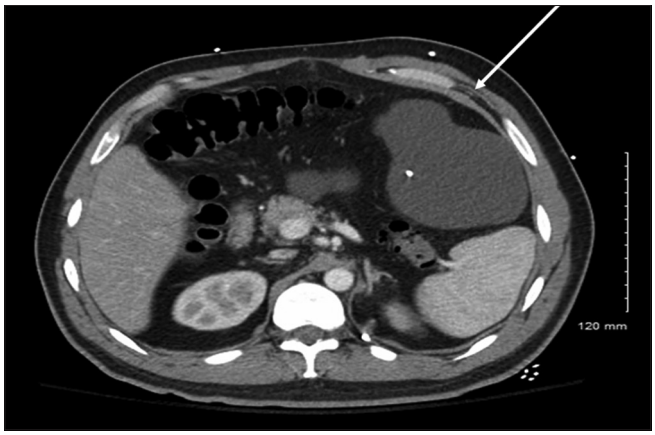

Figure 2: (Case #2) Computed tomography of the abdomen showing right upper quadrant abdominal pseudocyst (arrow) near the gastric outlet

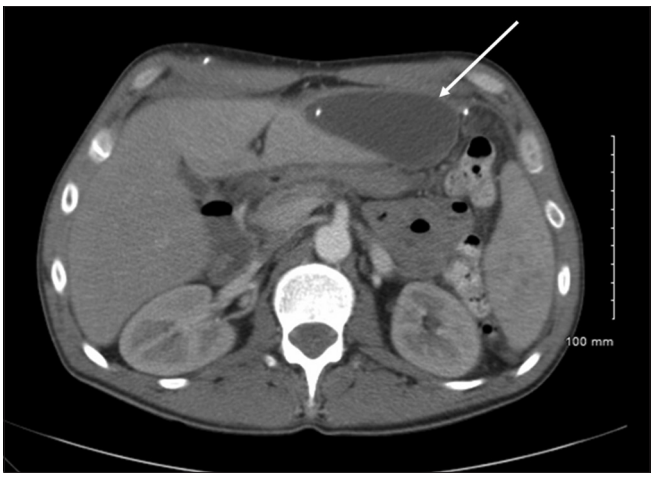

Figure 3: (Case #3) Computed tomography of the abdomen showing right upper quadrant abdominal pseudocyst (arrow)

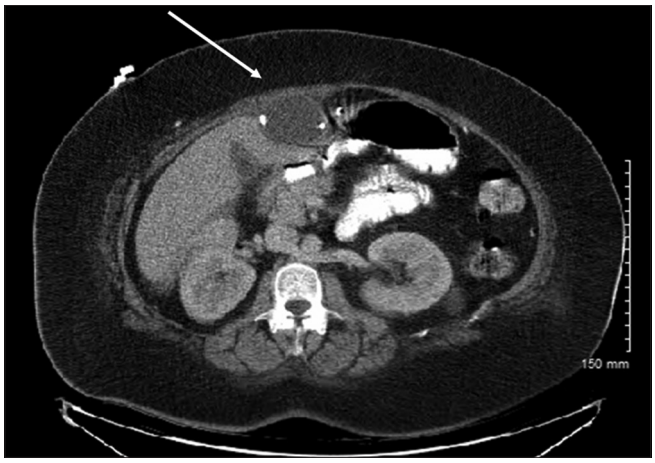

Figure 4: (Case #4) Pseudocyst (arrow) seen in the left anterior abdominal wall

Six years later, she was admitted to the hospital with sepsis, which was likely secondary to cellulitis as well as poorly controlled diabetes mellitus (A1C = 13.4%). She complained of pain on her abdominal wall, but otherwise had no neurological signs or symptoms. It was noted on admission that she had cellulitis along the tract of her distal shunt catheter. She was also noted to have pockets of underlying abscess in the soft tissue of her abdominal wall on a contrast CT abdomen. ESR on admission was 113; CRP was 22.05. Given the cellulitis secondary to the distal catheter, she underwent bedside externalization of the distal portion of her VP shunt. The culture from the wound, CSF, and catheter tip culture grew *Beta Streptococcus Group A*, necessitating an Infectious Disease consultation. An endoscopic third ventriculostomy was attempted, however, it could not be safely performed due to diffuse thickening of the floor of her third ventricle. After resolution of the soft tissue infection of her abdominal wall and infectious disease clearance, a new VP shunt was placed on the opposite side. A revision surgery was performed on the left side with the assistance of the general surgery service in laparoscopic placement of the distal catheter.

DISCUSSION

We have presented four cases of APC formation following VP shunt placement. These cases have informed our experience in the diversity of techniques that can be used to manage this complication. We propose an algorithm to select the best treatment option based upon the patient's presentation [Figure 5] using lessons, as illustrated in our previously discussed cases and in review of the literature.

The unique nature of our first case lies in the fact that the patient had an initial shunt revision performed at an outside hospital due to his abdominal pseudocyst causing impaired CSF drainage. The recurrence of APC is not necessarily uncommon, with the incidence in the literature varying widely from 7.1% to 62.5%.[7,12] What is unique in this case is that recurrence led to a second VP shunt malfunction and infection. It was our initial management decision to attempt a ventriculopleural shunt after 3 sets of negative CSF cultures, however, the patient was adamant that he wanted another attempt at placement of a peritoneal catheter despite being informed of the risks of another recurrence. This approach is not recommended as per our algorithm because pseudocyst formation is an indication that the peritoneum may not be useful for shunting, but it is ultimately the patient's decision.[1,10] Had the patient not desired a VP shunt, the preferred method of treatment would have been ventriculopleural shunt placement.

Our second case is especially rare because, to our knowledge, there have not been any cases of gastric outlet obstruction caused by APC reported in the literature. He was managed conservatively as he was asymptomatic in regards to neurological symptoms. If his pseudocyst was not causing gastric outlet obstruction, no intervention would have been recommended. Percutaneous drainage of the cyst to relieve the obstruction was the initial approach considered, however, given his pro-inflammatory state (ESR: 108, CRP: 14.30), there was a high index of suspicion that it would recur. In addition, it has been reported that there is a higher incidence in patients with prior abdominal surgery or history of necrotizing enterocolitis.[10] For this reason, his VP shunt was converted to a ventriculopleural shunt. This was follow by IR-guided drainage of the APC in addition to antibiotic therapy.

The third case demonstrates an example of APC as a low-grade inflammatory process and gives credence to the theory that these are secondary to a low-grade infection. It also demonstrates that a first time diagnosis of pseudocyst may be managed with simple percutaneous drainage and appropriate antibiotic treatment, if the CSF remains unaffected. It is important to note that, in addition to gram stain and culture of the APC aspirate, CSF studies should be obtained so that a VP shunt infection can be effectively ruled out.

Our final case reinforces the fact that VP shunts are prone to complications – with infection rates within the first month ranging 3–20%.[3,6,15,17] In addition, patients with poorly controlled diabetes mellitus are more prone to infections in general. In this patient's case, she had a number of revisions prior to her initial presentation to our service and she had multiple infections related to her shunt. Because her CSF culture and catheter culture grew *Beta Streptococcus Group A*, serial CSF cultures were drawn so that her shunt could be replaced. Because of her poorly controlled diabetes and multiple shunt malfunctions, an endoscopic third ventriculostomy (ETV)

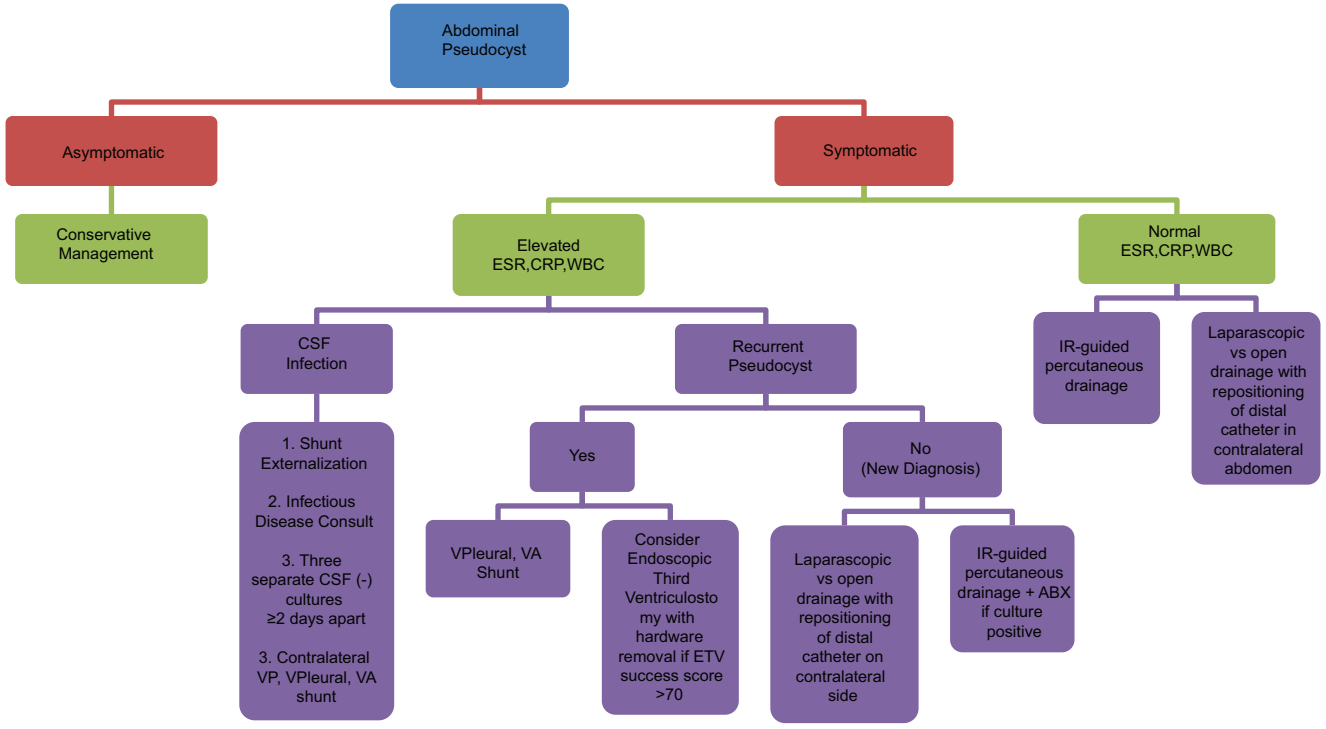

Figure 5: Proposed algorithm for decision making in management of abdominal pseudocyst secondary to ventriculoperitoneal shunt

was attempted in an effort to eliminate the possibility of future hardware infections. Her ETV success score was calculated to be 80. However, as stated previously, ETV could not be safely performed due to diffuse thickening of the floor of the third ventricle. Again in this case, replacement of hardware was not performed until infections of both soft tissue and CSF were clear on repeat cultures.

The classic signs of VP shunt malfunction are the same as the typical clinical manifestations of increased intracranial pressure, i.e., headache, nausea, vomiting, and altered level of consciousness. Abdominal complications related to VP shunt placement include pseudocysts (both infected and sterile), abscess, and bowel perforation. With regard to abdominal pseudocysts, the presentation can be more variable. Abdominal pain is often the chief complaint in most patients, accompanied by nausea and vomiting, which is often in the absence of neurological signs. CT scan is often more useful in distinguishing those presenting with severe abdominal pain because it can help identify other etiologies such as appendicitis, diverticulitis, abdominal abscess, or bowel obstruction. The diagnosis of pseudocysts is most accurately made by a contrast CT scan of the abdomen, although ultrasound has been recommended by some due to its frugality. Eliminating radiation exposure is an especially important consideration in the pediatric and pregnant population. The authors recommend CT scan of the abdomen with intravenous contrast as the preferred initial method of diagnosing abdominal pseudocysts, except in cases of pediatrics or pregnancy.

APC have been reported in the literature with the incidence ranging from 0.25% to 10%.[2,5,10,13,17] Some rare cases even result in intraaxial pseudocysts, with some cases having been reported to occur in the parenchyma of organs such as liver or spleen.[8] The etiology of pseudocysts is still unclear. They have been theorized to occur secondary to an inflammatory process, ranging from an infectious cause to a hypersensitivity reaction to the composition of the shunt catheter to malabsorption of CSF. Because of the potential for an underlying infection, the authors recommend obtaining CSF sample via shunt tap or lumbar puncture to send for gram stain and culture.

Treatment of pseudocysts is highly variable and there is no established standard, therefore, the method of treatment should be tailored to the overall clinical picture. Treatment options include percutaneous drainage of the pseudocyst with distal repositioning of peritoneal catheter (open or laparoscopic), placement of distal catheter in an alternative location such as the contralateral abdomen, the pleural space or the right atrium, and endoscopic third ventriculostomy with removal of shunt hardware completely.[9,10,13,18] Our proposed algorithm has been validated in the literature, with Mobley *et al*. advocating for an algorithmic approach to this condition.[10] A primary difference is advocating

for a conservative approach in asymptomatic patients. Moreover, we advocate for the use of ESR and CRP as the primary inflammatory markers as well as laparoscopic approaches for revision. In our cases, our proposed algorithm and all its approaches were followed, with the exception of the first case. In the first case, the option of VP shunt was discussed with the patient, however, he insisted on having the distal catheter repositioned in the left lower quadrant of his abdomen. This was due to the fact he was extremely anxious about a catheter being placed in his chest. In cases of such repeat operations, we recommend consulting a general surgeon for assistance so as to reduce the risk of further complication.

CONCLUSION

Abdominal pseudocysts are a known, but rare, complication of VP shunt placement. It is likely secondary to a low-grade inflammatory process. Diagnosis is best made by contrast CT scan of the abdomen except in childhood and pregnancy. CSF studies via shunt tap or lumbar puncture are also recommended to rule out hardware infection. In regards to the treatment algorithm, we propose placing an emphasis on the selection of alternative locations for shunt placement in symptomatic patients who are found to be in an inflammatory state, along with conservative management in asymptomatic patients. In our experience, this approach has resulted in a simplified approach to treatment in otherwise complicated patients.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

REFERENCES

1. Alduraibi S. Ventriculoperitoneal Shunt with Communicating Peritoneal & Subcutaneous Pseudocysts Formation. Int J Health Sci 2014;8.
2. Birbilis T, Kontogianidis K, Matis G, Theodoropoulou E, Efremidou E, Argyropoulou P. Intraperitoneal cerebrospinal fluid pseudocyst. A rare complication of ventriculoperitoneal shunt. Chirurgia 2008;103:351-3.
3. Browd SR, Gottfried ON, Ragel BT, Kestle JR. Failure of cerebrospinal fluid shunts: Part II: Overdrainage, loculation, and abdominal complications. Pediatr Neurol 2006;34:171-6.
4. Bryant M, Bremer A, Tepas 3rd J, Mollitt D, Nquyen T, Talbert J. Abdominal complications of ventriculoperitoneal shunts. Case reports and review of the literature. Am Surg 1988;54:50-5.
5. Burchianti M, Cantini R. Peritoneal cerebrospinal fluid pseudocysts: A complication of ventriculoperitoneal shunts. Child's Nerv Sys 1988;4:286-90.
6. Chung JJ, Yu JS, Kim JH, Nam SJ, Kim MJ. Intraabdominal complications secondary to ventriculoperitoneal shunts: CT findings and review of the literature. Am J Roentgenol 2009;193:1311-7.
7. Dabdoub CB, Dabdoub CF, Chavez M, Villarroel J, Ferrufino JL, Coimbra A, *et al.* Abdominal cerebrospinal fluid pseudocyst: A comparative analysis between children and adults. Child's Nerv Sys 2014;30:579-89.
8. Dabdoub CB, Fontoura EA, Santos EA, Romero PC, Diniz CA. Hepatic cerebrospinal fluid pseudocyst: A rare complication of ventriculoperitoneal shunt. Surg Neurol Int 2013;4.
9. de Oliveira RS, Barbosa A, Machado HR. An alternative approach for management of abdominal cerebrospinal fluid pseudocysts in children. Child's Nerv Sys 2007;23:85-90.
10. Doran S, Hellbusch L. Abdominal pseudocyst: Predisposing factors and treatment algorithm. Pediatr Neurosurg 2005;41:77-83.
11. Kausch W. Die behandlung des hydrocephalus der kleinen Kinder. Arch Klin Chir 1908;87:709-96.
12. Laurent P, Hennecker J, Schillaci A, Scordidis V. [Abdominal CSF pseudocyst recurrence in a 14-year-old patient with ventricular-peritoneal shunt]. Arch Pediatr 2014;21:869-72.
13. Moussa WMM, Mohamed MAA. Efficacy of postoperative antibiotic injection in and around ventriculoperitoneal shunt in reduction of shunt infection: A randomized controlled trial. Clin Neurol Neurosurg 2016;143:144-9.
14. Pernas JC, Catala J. Case 72: Pseudocyst around Ventriculoperitoneal Shunt 1. Radiology 2004;232:239-43.
15. Rainov N, Schobess A, Heidecke V, Burkert W. Abdominal CSF pseudocysts in patients with ventriculo-peritoneal shunts. Report of fourteen cases and review of the literature. Acta Neurochir 1994;127:73-8.
16. Sharma AK, Pandey AK, Diyora D, Mamidanna R, Sayal P, Ingale HA. Abdominal CSF pseudocyst in a pateint with ventriculoperitoneal shunt. Indian J Sur 2004;66. [misspelling of 'pateint' is how the article is published].
17. Tamura A, Shida D, Tsutsumi K. Abdominal cerebrospinal fluid pseudocyst occurring 21 years after ventriculoperitoneal shunt placement: A case report. BMC Surg 2013;13:1.
18. Yuh SJ, Vassilyadi M. Management of abdominal pseudocyst in shunt-dependent hydrocephalus. Surg Neurol Int 2012;3:146.