Case Report

A Rare Abdominal Complication of Ventriculoperitoneal Shunt: Hepatic Cerebrospinal Fluid Pseudocyst

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A B S T R A C T

Introduction: Ventriculoperitoneal shunt (VP) is one of the most commonly performed procedure in the management of hydrocephalus. For the absorption of cerebrospinal fluid (CSF), abdominal cavity has been always considered as a good option. Approximately 0.25- 40% cases may present with complications, post VP shunt operation. Abdominal CSF pseudocyst is rare complication.

Case report: A 14 years/Male was admitted to the hospital with chief complaints of abdominal pain located at right upper quadrant since 10 days, with past history of suboccipital craniotomy for total excision of pilocytic astrocytoma with VP shunt, 2 years earlier. Laboratory studies showed mild elevation of SGOT and SGPT values. Imaging studies demonstrated a thick walled cystic lesion in the sub capsular region of couinaud segment VI of liver; with the tip of VP shunt catheter seen just medial to the collection. No organisms were recovered in the sample, obtained through USG guided aspiration of collection. Post-aspiration, sub capsular hepatic CSF pseudocyst appears collapsed and patient started improving symptomatically. Follow up ultrasound was performed and complete resolution of cyst was noted.

Conclusion: Abdominal CSF pseudocyst is a uncommon complication after VP shunt operation and may lead to shunt malfunction or abdominal signs/symptoms. If suspicion of CSF abdominal cyst is raised, it should be confirmed by imaging. Repositioning/removal of VP shunt catheter or simple aspiration of cyst can be done as part of treatment protocol.

Keywords: CSF Pseudocyst, Hepatic Pseudocyst, Ventriculoperitoneal Shunt

INTRODUCTION

Peritoneal cavity is considered best site for CSF absorption; so ventriculoperitoneal shunt is gold standard for the management of hydrocephalus. Its complications include obstruction of proximal or distal catheter tip, peritonitis, cerebrospinal fluid ascites, infection, abdominal CSF pseudo cyst (rare). Pseudo cyst formation is common in pediatric population[1] because of more incidence of hydrocephalus in pediatric population. Some of the pseudo cysts are benign and remain asymptomatic; whereas some may lead to potential abdominal pain and even malfunction of shunt system.This case report describes a case of extra-axial hepatic CSF pseudo cyst, status post op VP shunt operation and presents the clinical scenario, radiological findings and surgical management.

CASE REPORT

A 14 years old boy presented with chief complaints of abdominal pain located at right upper quadrant , dull aching type of pain radiating to right shoulder , which has been progressing for the past 10 days. Pain aggravates on food intake and there is no relieving factor. His past surgical history includes VP shunt placement following total resection of pilocytic astrocytoma through midline sub occipital approach craniotomy, done in 2018. Initial inspection revealed, midline inverted umbilicus, midline scar present (VP shunt), no swelling sinus or dilated veins, no abnormal pulsation or peristalsis. On palpation, mild tenderness was present at right hypochondriac region, liver was palpable 2 cm below costal margin. On percussion, no evidence of free fluid and bowel sounds appear normal on auscultation. Cardiovascular, respiratory and central nervous system examination were within normal limits.

Blood sample was sent for hematological examination and showed SGOP-55 U/L, SGPT-70 U/L, alkaline phosphatase-255 U/L, total bilirubin-0.3 mg/dl, indirect bilirubin-0.2 mg/dl, direct bilirubin-0.1 mg/dl, total counts-12,200 cells/cumm, Hb-11 g/dl, INR-1, urea-14 mg/dl, creatinine-0.6 mg/dl, random glucose-96 mg/dl, sodium-137 mEq/L, potassium-4.4 mEq/L , Chloride -105 mEq/L.

USG abdomen and pelvis was performed and it revealed an anechoic cystic lesion (Fig. 1) (vol~ 36cc), with numerous echogenic septations within; located in couinaud segment VI of right lobe of liver. No obvious solid component was noted within. Cystic lesion showed no evidence of internal vascularity on color doppler (Fig. 2). Abdominal end of VP

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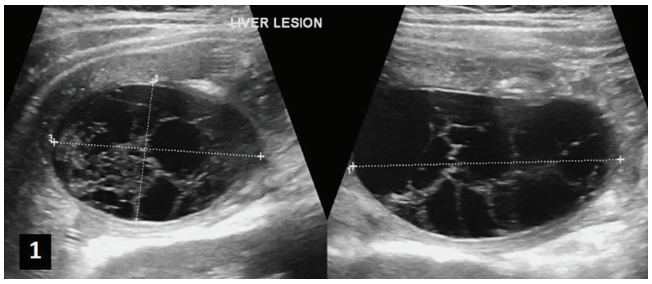


Figure-1: Transverse and sagittal ultrasound image of an anechoic cystic lesion, measuring~36 cc in volume, with numerous echogenic septations within, located at couinaud segment VI of right lobe of liver.

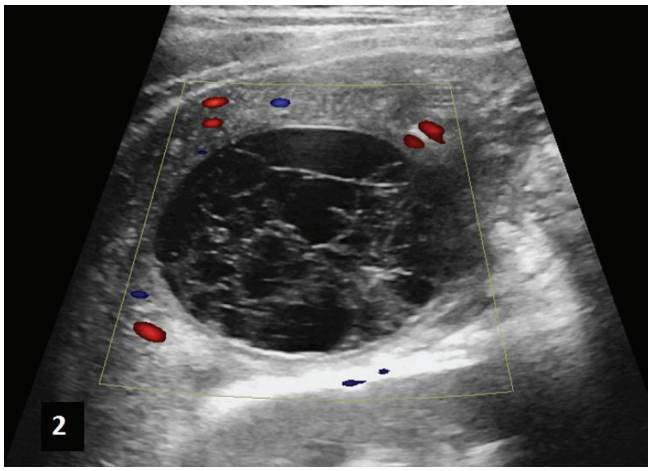


Figure-2: Transverse ultrasound image of an anechoic cystic lesion show no evidence of internal vascularity on colour doppler.

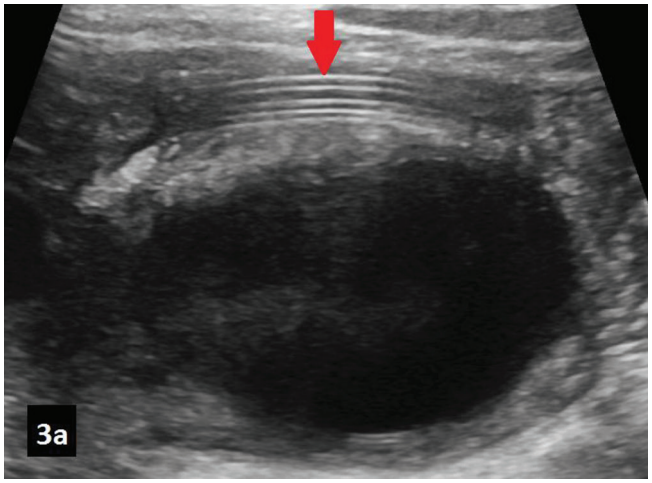


Figure-3a: Ultrasound image showing a parallel echogenic stripe- 'rail-road sign' representing abdominal end of VP shunt (red arrow), encircling the cyst .

shunt was noted in close proximity to the lesion (Fig. 3a, 3b). NCCT brain was performed and it showed proximal tip of VP shunt in situ in III ventricle (Fig. 4). CECT abdomen showed a thick walled (measuring ~0.4 cm) cystic lesion in sub capsular region of segment VI of the right lobe of liver (Fig. 5a,5b). Walls of cyst were intact. The tip of VP shunt was seen just medial to the cystic lesion (Fig. 6,7). Under USG guidance diagnostic and therapeutic aspiration

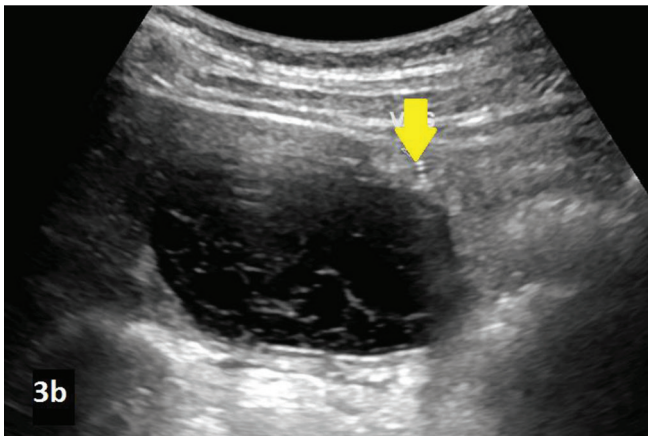


Figure-3b: Ultrasound image showing tip of abdominal end of VP shunt (yellow arrow) in close proximity to cystic lesion.

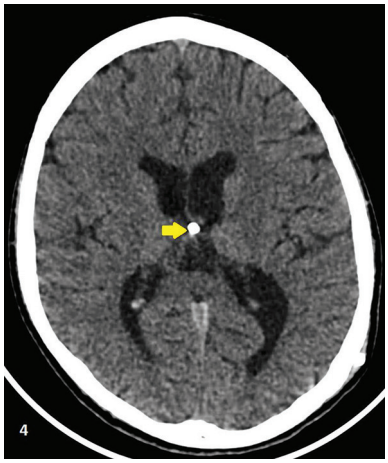


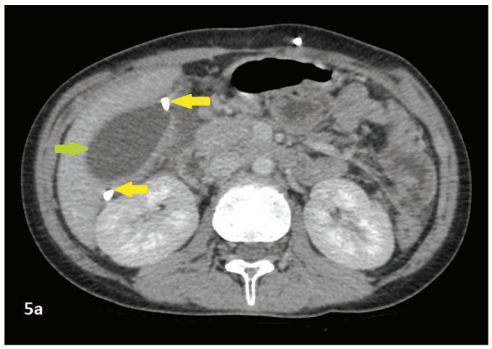
Figure-4: Axial image of NCCT brain taken at the level of third ventricle, shows cranial tip of VP shunt (yellow arrow) in III ventricle

was performed. Approximately 32 ml of fluid was aspirated and sent for cytology. Cyst appears to be collapsed after ultrasound guided aspiration(Fig. 8). Cytology report of aspirated sample revealed clear fluid, with no evidence of organism within. Biochemical examination of fluid revealed picture similar to cerebrospinal fluid. Culture report of the sent sample was negative. On fourth day of hospitalization, patient was discharged with complete resolution of symptoms.

DISCUSSION

According to international literature, abdominal CSF pseudocyst accounts for about 6.8%[2] of all VP shunt complications. Otherwise, a hepatic pseudocyst secondary to VP shunt is a very rare complication. Incidence of abdominal CSF pseudocyst is quite high in pediatric patient as compared to adult, because hydrocephalus is more common in children. Rainvo et al[3] reported a child adult ratio of 1.8:1.

Hepatic CSF pseudo cyst can be (1)Intra-axial type, when the tip of VP shunt lodge into the liver parenchyma and can cause intra-axial pseudo cyst formation (2) Extra-axial type, when the tip of catheter penetrate Glisson's capsule leading to sub capsular pseudocyst formation or (3) Juxtahepatic CSF pseudo cyst, in which small part of cyst is within the hepatic parenchyma with the large part lying outside.



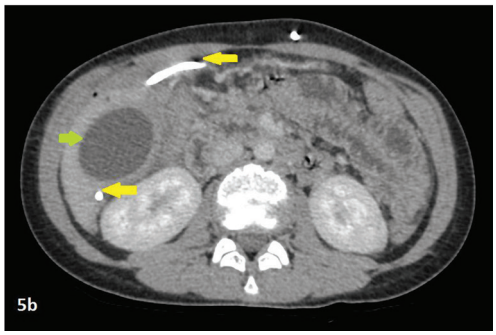


Figure-5a,5b: Axial sections of CECT abdomen at venous phase shows a well defined non-enhancing cystic lesion (green arrow) in couinaud segment VI of right lobe of liver; with abdominal end of VP shunt catheter (yellow arrow) encircling the cyst.

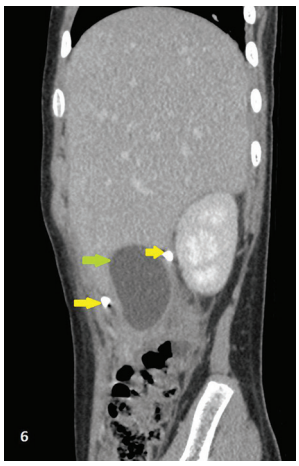


Figure-6: Sagittal sections of CECT abdomen at venous phase shows VP shunt (yellow arrow) entering the hepatic capsule and entering into the cystic lesion (green arrow).

Although the etiology of abdominal CSF pseudo cyst is not clear[1], different authors believe inflammatoryprocess, either sterile or infectious; as predisposing factor. Other predisposing factors can be (1) past abdominal surgeries or repeated shunt reversal surgeries, (2) defect in absorption of CSF, (3) increase in protein content of CSF and (4) allergic reaction to silicon or ethylene oxide. All the mentioned factors are capable of formation of peritoneal adhesions, that cause prevention of CSF reabsorption and can lead to cyst formation. Consequently, oncotic pressure of cystic fluid increases, more interstitial fluid enters into the pseudo cyst; causing increase in cyst size. Formation of abdominal CSF pseudo cyst takes few days to several years , after VP shunt

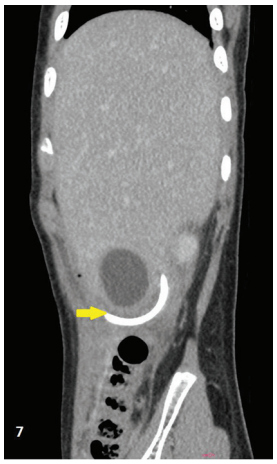


Figure-7: Coronal section of CECT abdomen at venous phase shows a well defined non-enhancing lesion noted at couinaud segment VI of liver. VP shunt (yellow arrow) .

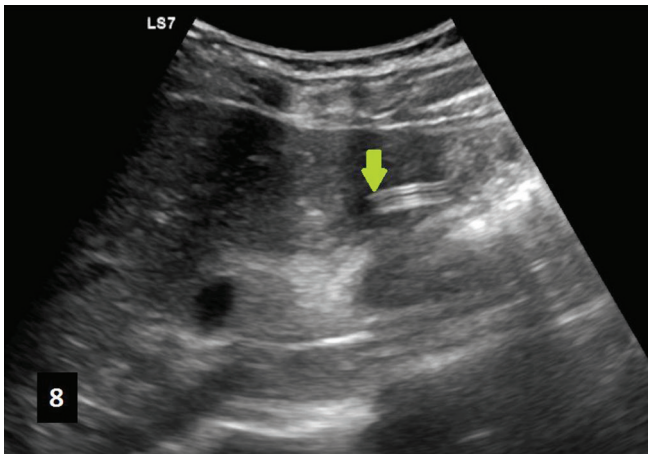


Figure-8: Post aspiration follow up ultrasonography reveals complete collapse (green arrow) of subcapsular hepatic CSF pseudocyst.

implantation. The most important factor predisposing to CSF pseudo cyst is malposition of peritoneal tip of shunt to the liver surface, causing chronic irritation and focal hepatic injury. According to mobley et al[4], most common cause of hydrocephalus for which shunt has to be placed is intra-ventricular hemorrhage, congenital hydrocephalus, dandy walker cyst and myelomeningocele. In patients of infected abdominal CSF pseudo cyst, most common organism isolated were S.epidermidis and S.aureus.

Abdominal CSF pseudo cyst does not have any pathogonomic[5] signs but most common presentations are abdominal pain, abdominal distention, palpable mass, constipation due to pressure effect, intestinal obstruction, nausea or vomiting.After days and weeks of abdominal symptoms, CNS symptoms may start. This delay in onset of CNS symptom is due to continued slow absorption of CSF, although inadequate to prevent CSF pseudo cyst formation. The confirmed diagnosis of hepatic CSF pseudo cyst cannot be made on laboratory examinations alone. Several patients with this condition may show elevated levels of SGOT, SGPT and alkaline phosphatase. These deranged values points out the existence of anicteric cholestasis along with

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hepatic cell necrosis.

On radiological imaging ultrasound holds good position because it is non invasive, easy to perform, inexpensive and can give a satisfactory diagnosis. Imaging findings include migration of tip of the peritoneal end of catheter into a large fluid filled collection . The double echo of the shunt catheter wall may produce the so-called 'rail road sign'. Abdominal CECT scan can provide a more accurate diagnosis. In extra-axial pseudocyst, hepatic tissue cannot be identified in the peripheral wall of pseudocyst.CT scan withhold the first choice of investigation as it can exclude abdominal abscess, appendicitis, volvulus or diverticulitis.

Treatment of abdominal CSF pseudocyst depends on the patient characteristic. The experience of surgeon[6] and the findings during the operation. After the appropriate treatment of infection, if present; one among these procedure[7] can be done- (1) Repositioning of distal end of catheter into the opposite site of the peritoneum or into the non peritoneal space (atrium, gall bladder, pleural space), (2) Exploratory laparotomy with lysis of adhesions, followed by repositioning of catheter, (3) Laparoscopic drainage and repositioning of catheter tip, (4) Simple aspiration of abdominal CSF pseudocyst and (5) Shunt removal or disconnection. In presence of infection, external drainage has to be done, along with treatment of infection.

In our case, subcapsular hepatic CSF pseudocyst was treated[8] by USG guided cyst aspiration. In our case there was no evidence of infection and the cyst was extra-axial. So, once we verified the absence of infection; definite treatment was done. Care has to be taken while, aspiration of hepatic CSF pseudocyst because of the risk of bleeding .

Sometimes abdominal CSF pseudo cyst can recur. Ersatin et al[9] reported 20% of recurrence. Case was followed up for period of 2 years, and no residual/recurrence of hepatic CSF pseudocyst was evident on ultrasound.

CONCLUSION

Abdominal CSF pseudo cyst is a uncommon complication after VP shunt operation and may lead to shunt malfunction or abdominal signs/symptoms. If suspicion of CSF abdominal cyst is raised, it should be confirmed by imaging. Repositioning/removal of VP shunt catheter or simple aspiration of cyst can be done as part of treatment protocol, according to patient presentation and clinical symptoms.

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