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Case Report

A rare complication of ventriculoperitoneal shunt: Pleural effusion without intrathoracic ventriculoperitoneal shunt catheter

Said Hilmani[1], Tarek Mesbahi[1], Abderrahman Bouaggad[2], Abdelhakim Lakhdar[1]

[1]Department of Neurosurgical, SAID Hilmani, [2]Department of Anesthesiology, Clinic Dar Salam, UHC Ibn Rusch, Casablanca, Morocco.

E-mail: *Said Hilmani - hilmani.said@yahoo.fr; Tarek Mesbahi - dr.t.mesbahi@gmail.com; Abderrahman Bouaggad - darsalam.bouaggad@gmail.com; Abdelhakim Lakhdar - abdelhakim.lakhdar@yahoo.com

***Corresponding author:**
Said Hilmani,
Department of Neurosurgical,
SAID Hilmani, Casablanca,
Morocco.

hilmani.said@yahoo.fr

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ABSTRACT

Background: Symptomatic pleural effusion following ventriculoperitoneal shunt (VPS) insertion is very rare and poorly understood in the literature in contrary to other mechanical complications.

Case Description: We report a case of 15 month-year-old girl who had VP shunt for congenital hydrocephalus. Twelve months after surgery, she was diagnosed with massive hydrothorax. Chest X-ray and thoracoabdominal CT scan confirmed the right pleurisy and showed the tip of the peritoneal catheter in the general peritoneal cavity. We made thoracic drainage of the transudative pleural effusion. When we released the chest tube, 24 h after, the girl showed a respiratory distress again and the effusion resumed at the X-ray control. Her symptoms abated after the realization of a ventriculoatrial shunt "VAS." Repeat chest X-ray confirmed the resolution of the hydrothorax.

Conclusion: Despite the not yet well-understood mechanism of this rare and important VPS complication, management is simple based on X-ray confirmation, thoracentesis with biological analysis, and catheter replacement, especially in atrium "VAS."

Keywords: Hydrothorax, Ventriculoatrial shunt, Ventriculoperitoneal shunt complication

INTRODUCTION

Mechanical and shunt infection are the most common complications of ventriculoperitoneal shunt (VPS), especially in the pediatric patients treated for hydrocephalus. Pleural effusion complicating VPS is a very rare condition. Most cases described with hydrothorax are due to the migration of the catheter tip into the pleural space.[2,5,16] There are few published cases with a normopositioned shunt catheter. Most of the reported cases occur in children.[1,4] In this study, we report 15 months old infant with pleural effusion following VP shunt for congenital hydrocephalus without catheter migration and explain the pathophysiology.

CASE REPORT

A 15-month-old girl presenting a congenital hydrocephalus, diagnosed at uterine life. At the age of 3 months, a right VAS was made due to the progressive character of her congenital hydrocephalus.

Immediate and short time follow-up was simple. One year later, the infant goes to the emergency department for a progressive respiratory distress with polypnea and diminution of oxygen saturation. She was afebrile with pulmonary auscultation evidencing hypophony of the right hemithorax and abdomen not distended and depressible without pain. The blood analysis did not show significant alterations. A thoracoabdominal X-ray shows complete opacity of the right hemithorax, without evidence of intestinal obstruction or intraperitoneal air and with a well-positioned catheter tip but in contact of diaphragm on sleep position and in pelvic cavity on up position [Figure 1]. This was confirmed by ultrasound exploration without CSF ascites. A thoracoabdominal scan revealed the right pleural hydrothorax and the position of the catheter tip in the peritoneal cavity [Figure 2]. The patient has been transferred to the pediatric intensive care unit and right pleural effusion drainage has been performed, obtaining a clear crystal appearance and transudative fluid. Proteinorachia and glycorachia were normal in CSF microbiological studies and culture excluded infection. Noninvasive ventilatory support is initiated with oxygen therapy. We had progressive clinical improvement after cessation of drainage debit and the radiographic disappearance of the effusion after 48 h. A cardiac cause was eliminated by a strictly normal cardiac examination and echocardiography not compatible with heart failure, as well as a nephrological origin by the biological analyzes. However, clamping of the chest tube was marked by recurrent pleural effusion 24h after with polypnea. A ventriculoatrial shunt was performed with simple operative follow-ups. Clinically, the patient became normal and resolution of the pleural effusion was obtained on the thoracoabdominal control X-ray [Figure 3].

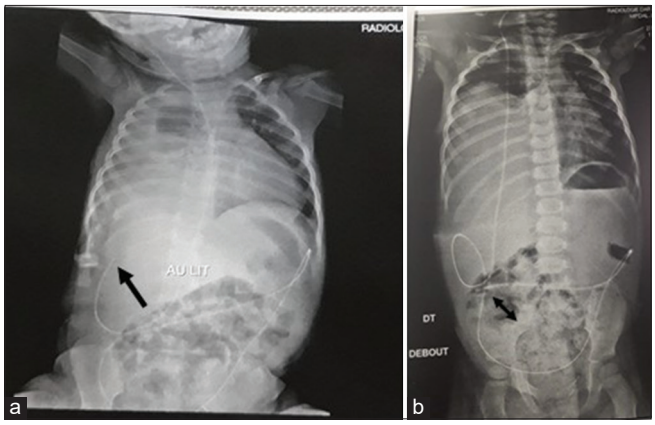

Figure 1: (a) Sleep chest and abdomen radiograph demonstrates that complete opacity in right hemithorax with cather tip is in contact of diaphrragm "black arrow," (b) the tip catheter is in peritoneal cavity on up postion. "Double black arrow."

DISCUSSION

Mechanical failures and infections are the common complications of VP shunts.[9] Mechanical failure is the most frequent cause of shunt malfunction occurring during the first 2 years after shunt placement with more than 40%.[7,9] Malfunction due to infection occurs approximately in 5–15%.[5] CSF pleural effusion in VP shunted patients is a rare complication. Since 1977 and at our knowledge, 26 cases are reported in the literature and most frequently associated with distal catheter tip migration into the thorax secondary to intrathoracic trauma during the placement in over 60% of cases.[2] However, cases without catheter migration into the thorax still very rare [Table 1].[8,11,18] They are predominantly described in the pediatric population and on the right side.[3,6,19] The mechanism of pleural effusion is comprehensible in iatrogenic communication with the pleural space and secondary to an intrathoracic catheter migration.[17] In patients with normal placement of VP shunt in the peritoneal cavity, the pathophysiology is relatively

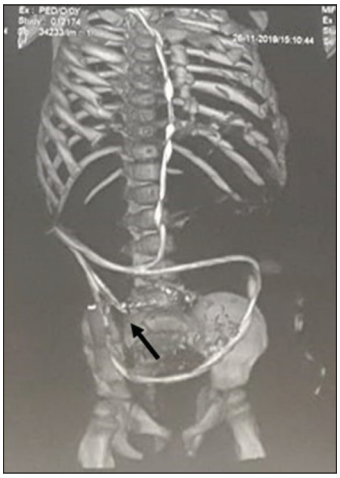

Figure 2: Thoracoabdominal CT scan confirms catheter tip in peritoneal cavity. "Black arrow."

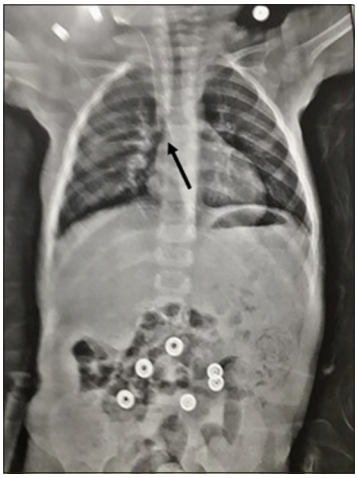

Figure 3: Postoperative chest X-ray view shows presence of shunt in the right atrium "black arrow" with disappearance of pleurisy.

Table 1: Series of CSF hydrothorax in children without intrathoracic catheter migration.

Authors/year	Age	Delay from VPS	Ascitis	Treatment
Glöbl and Kaufman, 1978[7]	No info	No info	+	No info
Faillace *et al*., 1998[6]	4 months	1 months	-	Thoracentesis and VA positioning
Hadzikaric *et al*., 2002[8]	16 months	2 months	-	Thoracentesis and VA derivation positioning
Adeolu *et al*., 2006[1]	8 years	2.5 months		Shunt review
Born *et al*., 2008[3]	2.5 years	1.5 years	+	Thoracentesis and shunt review
Smith *et al*., 2009[16]	14 months	2.5 months	+	Thoracentesis and shunt externalization
Kocaogullar *et al*., 2011[11]	5 years	4.7 years	-	Thoracentesis and VA positioning
Patel *et al.,* 2011[14]	6 months	1.5 months	-	Thoracentesis, shunt review, and VA derivation positioning
Chuen-im *et al*., 2012[4]	5 years	5 years	-	Thoracentesis, pleurodesis, and intracranial endoscopic choroid plexus coagulation
Ulus *et al.,* 2012[19]	2 weeks	6,5 months	-	Thoracentesis and VA positioning
O'Halloran *et al*., 2013 [13]	5 years	2 years	-	Thoracentesis, shunt review, and VA derivation positioning
Kim *et al.,* 2015[15]	3 years	3 years	-	Thoracentesis VP shunt repositioning
Yéboles *et al*., 2017[20]	30 months	11 months	-	Thoracentesis
Tirado *et al*., 2019[18]	13 months	4 months	-	Thoracentesis, elevation of pressure from 60 mm H_2O to 190 mm H_2O (adjustable valve). Opening valve pressure was gradually reduced to 60 mm H_2O during 3 weeks

unclear and raises a lot of hypotheses. It is estimated in the literature that it could be secondary to leak of CSF circulating pericatheter and passing to the thoracic cavity or pleural effusion through congenital diaphragm continuity solutions, such as the Morgagni foramen and/or Bochdalek foramen.[17] This is a consequence of the increase in abdominal pressure secondary to CSF ascites which is not frequently reported, where CSF accumulates in the peritoneal cavity as a result of defective absorption.[10,12,13] Furthermore, authors hypothesize that local inflammatory reactions or repeated microtrauma induced by the shunt tip may contribute to the diaphragm erosion facilitating CSF effusion, as showed in our patient.[14,20] The negative intrathoracic pressure and the positive intra-abdominal pressure contribute to the fluid shift.[15]

In our case, the pleural effusion is right, ipsilateral to the position of the VP shunt, in which as the majority of cases described in the literature. The imaging allowed us to confirm the existence of the distal tip of the catheter in intra-abdominal and showed a minimum amount of free peritoneal fluid, with no clear ascites. The cause of CSF passage from the abdominal cavity to the thorax is not clear. We suggest that diaphragmatic catheter tip microtrauma is the most cause. However, we cannot rule out the existence of a continuity diaphragm solution. Finally, there are still questions that remain unanswered. Why hydrothorax occurs on the right side, without concomitant CSF ascites and many months or years after VPS in the majority of cases?

CONCLUSION

Hydrothorax following a VPS without catheter migration is an uncommon and serious complication. Contrary to the unclear mechanism, management is simple based on imaging, thoracentesis with biologic analysis, and shunt revisions. Different types of CSF shunting (VA shunt) or endoscopic treatment (third ventriculostomy with choroid plexus coagulation) may be considered as alternative therapeutic approaches.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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