Recurrent Symptomatic Pleural Effusion Due to a Ventriculopleural Shunt

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Ventriculopleural shunts are uncommonly used for the treatment of normal pressure hydrocephalus in adults. Pleural effusion has been reported to complicate the course of these ventriculopleural shunts in children. The pleural effusion should typically resemble the cerebrospinal fluid unless frankly infected. There are few good data on the nature of the pleural effusion. We, report a case of recurrent right-sided pleural effusion, 2 years after a ventriculopleural shunt insertion, for normal pressure hydrocephalus with no evidence of an underlying infection. The effusion abated after ligation of the shunt. We discuss the possible mechanisms in the development of the effusion. It is important to be aware of this unlikely complication of an uncommon procedure. Recognizing the origin of the pleural effusion can help in instituting close follow-up and early referral for revision of the ventriculopleural shunt. *Key words: ventriculopleural shunt, pleural effusion, exudates, recurrence*. [Respir Care 2009;54(8):1112–1114. © 2009 Daedalus Enterprises]

Introduction

The pleural space can be used as an alternative reservoir for diverting the cerebrospinal fluid in management of hydrocephalus,[1,2] especially if the peritoneal cavity is suboptimal for any reason. Pleural effusions, shunt obstruction, pneumothorax, and empyema have complicated the course of these shunts,[2-4] but have been infrequent. Most reports of pleural effusions complicating the course of ventriculopleural shunts have been in young adults and children. Data on the nature of these effusions are limited.

Case Report

A 59-year-old man presented with progressive dyspnea, fatigue, fever, and diarrhea for 2 days. He had a history of long-term cognitive impairment and epilepsy, well controlled with medications. Two years previously a ventriculopleural shunt was inserted for normal pressure hydrocephalus, with an initial valve pressure of 140 cm H_2O.

He had been admitted on 3 occasions in the previous 3 months, with worsening dyspnea. Chest radiographs revealed recurrent moderate right pleural effusion. On each occasion, therapeutic thoracentesis with drainage of approximately 1.5 L of pleural fluid provided symptom relief. The intervals between hospitalizations had decreased from 4 weeks initially to less than 2 weeks on subsequent occasions. Pleural fluid characteristics on each encounter, including the present admission, are summarized in Table 1. All samples had elevated lactate dehydrogenase levels and lymphocytosis.

SEE THE RELATED EDITORIAL ON PAGE 1026

On this admission, he was dehydrated and febrile, with tachypnea at rest. There was no cyanosis or palpable lymphadenopathy. The right chest was dull to percussion, with absent air entry up to the midzone. Normal heart sounds, with no murmurs or rubs, were noted on cardiovascular examination. The rest of the systemic examination was unremarkable. Laboratory examination revealed a hemoglobin of 10.1 g/dL (normal range 13.9–16.3 g/dL), with normal thyroid and renal functions tests and no leukocytosis. An echocardiogram was normal. A right pleural ef-

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Table 1. Pleural Fluid and Serum Values on Each Admission.

	Admission 1		Admission 2		Admission 3		Current Admission		
	Pleural Fluid	Serum	Pleural Fluid	Serum	Pleural Fluid	Serum	Pleural Fluid	Serum	CSF
Amount drained (L)	2	NA	1.4	NA	1.3	NA	1.5	NA	NA
pH	7.45	ND	7.39	ND	8.0	ND	7.44	ND	ND
Total protein (g/dL)	0.6 (normal 0.6–4.9)	6.6 (normal 6–8)	2.1	5.5	1.5	4.9	1.5	5.1	230 mg/dL (normal 20–45)
Lactate dehydrogenase (units/L)	90	97 (normal 98–192)	110	142	97	103	109	112	NA
Albumin (g/dL)	<1.0	2.3 (normal 3.1–4.7)	<1.0	1.7	<1.0	1.9	ND	ND	ND
Cholesterol (mg/dL)	29	ND	26	ND		ND	ND	ND	ND
White blood cells (cells/μL)	86	ND	13	ND	231	ND	127	ND	2
Lymphocytes (%)	60	ND	86	ND	86	ND	91	ND	11
Red blood cells (cells/μL)	30	ND	49	ND	43	ND	53	ND	5,000

CSF = cerebrospinal fluid
NA = not applicable
ND = no data collected

fusion was noted on the chest radiograph. A computed tomogram scan of the chest confirmed a large right pleural effusion with right-lower and middle-lobe atelectasis, and a ventriculopleural shunt in the lower-right hemithorax (Fig. 1). There was no evidence of pleural thickening, loculations, or masses.

A diagnostic and therapeutic thoracentesis was undertaken, draining 1,500 mL of clear fluid with a pH of 7.44, a total protein of 1.5 g/dL (normal range 0.6–4.9 g/dL), lactate dehydrogenase of 109 units/L, and a white-blood-cell count of 166 cells/μL, with 91% lymphocytes. Serum total protein and lactate dehydrogenase were 5.1 g/dL (normal range 6–8 g/dL) and 112 IU/L (normal range 98–192 IU/L), respectively. The pleural fluid was positive for β_2 transferrin, indicating the presence of cerebrospinal fluid. A simultaneous lumbar puncture was undertaken to rule out an infected shunt system. It revealed normal cerebrospinal fluid characteristics. The pleural and cerebrospinal fluid cultures were negative.

Stool studies confirmed *Clostridium difficile* colitis that was appropriately treated, with resolution of diarrhea and fever. A shunt series indicated that the tip of the catheter

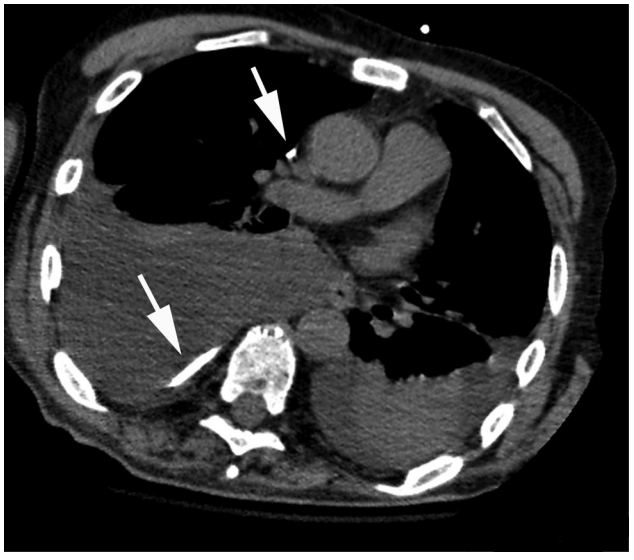

Fig. 1. Computed tomogram showing predominant right pleural effusion with the ventriculopleural shunt catheter in the right hemithorax (arrows).

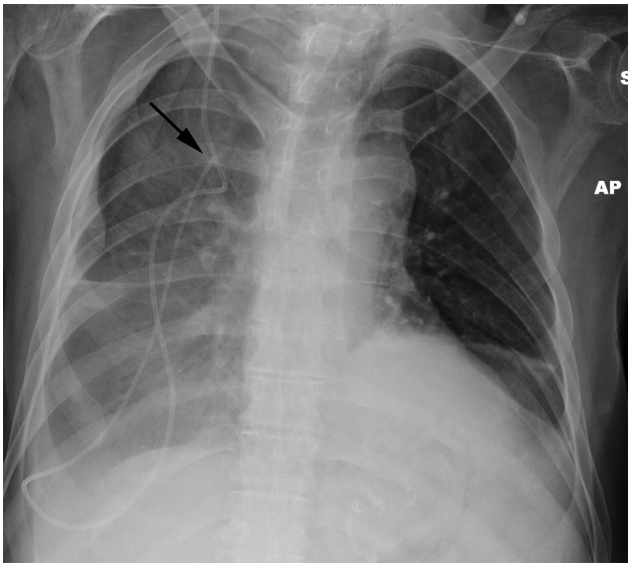

Fig. 2. Ventriculopleural shunt series revealing the tip of the catheter in the right pleural space (arrow), with moderate right pleural effusion.

was in the right pleural space (Fig. 2). The distal shunt catheter was ligated, as the patient declined a revision surgery. No symptomatic re-accumulation of the pleural effusion was noted after 6 months of follow-up.

Discussion

Ventriculopleural shunts were introduced as a management option for hydrocephalus by Ransohoff in 1954.[1] The next few decades witnessed reports of pleural effusions complicating the course of ventriculopleural shunts in infants.[2,3] This led to the relegation of ventriculopleural shunting as a "backup" measure. Some reports suggested that these shunts should preferably be avoided in younger children.[3] Incorporation of anti-siphon devices and valves into the shunt system has been recommended in an attempt to reduce the incidence of pleural effusions.[4]

Ventriculopleural shunts are rarely utilized in middle-age adults.[5] Despite the good absorptive capacity of the pleura in adults, some patients can develop pleural effusions.[5] The pleural fluid typically resembles the cerebrospinal fluid, with low protein and lactate dehydrogenase values.[6]

In our patient, the diversion of the cerebrospinal fluid into the pleural space was utilized as a first-line measure, due to his abdominal size. Recurrent pleural effusions developed around 2 years after the shunt insertion. The pleural fluid consistently demonstrated high lactate dehydrogenase levels and lymphocytosis. No further re-accumulation of the effusions was noted after ligation of the shunt. His fever on the current admission was due to *Clostridium difficile* infection, which responded to appropriate antibiotic treatment.

Pleural effusions develop due to the alterations in dynamics of net pleural fluid production and absorption. The mechanisms for accumulation of pleural effusions with ventriculopleural shunts remain speculative. The presence of a shunt catheter in the pleural space may produce local irritant effects, inducing a chronic sub-clinical inflammatory response, as supported by the predominant lymphocytosis in the pleural fluid. Inflammation leads to increased pleural fluid production and impaired lymphatic flow, causing pleural fluid accumulation and lung collapse. This further reduces the pleural surface area, resulting in a decrease in the net absorption of the pleural fluid.[7] Continuous addition of cerebrospinal fluid compounds the problem, leading to rapid accumulation of large pleural effusions.[7]

The elevated pleural fluid lactate dehydrogenase could be due to chronic shunt catheter inflammation, and partly as result of repeated thoracentesis causing cell injury.[8] The possibility of a local immune response cannot be excluded.[5] In adults there is probably a gradual diminution in the pleural resorptive capacity over time; hence, symptoms may not manifest for several months.

The presence of β_2 transferrin in a body fluid has 94% sensitivity and almost 100% specificity for the presence of cerebrospinal fluid.[9] Hence, it should be considered in all patients with a ventricular shunt and a pleural effusion.

Some degree of pleural effusion is noted in most patients with ventriculopleural shunts. Asymptomatic or minimally symptomatic effusions mandate close follow-up, with elective referral to the neurosurgeon. Therapeutic thoracentesis can be utilized as a temporizing measure for large symptomatic effusions. Revision of the shunt remains definitive treatment.[10]

In conclusion, ventriculopleural shunt insertion, though uncommon, is occasionally utilized in adults. Clinicians need to be alert to the possibility of pleural effusions complicating the course of these ventriculopleural shunts, and that those could present a few years later. They could be transudative or exudative in character.[6] Revision of the shunt may be considered in these cases.

REFERENCES

1. Ransohoff J. Ventriculo-pleural anastomosis in treatment of midline obstructional neoplasms. J Neurosurg 1954;11(3):295-298.
2. Nixon HH. Ventriculo-pleural drainage with a valve. Dev Med Child Neurol 1962;4(6):301-302.
3. Hoffman HJ, Hendrick EB, Humphreys RP. Experience with ventriculo-pleural shunts. Childs Brain 1983;10(6):404-413.
4. Martinez-Lage JF, Torres J, Campillo H, Sanchez-del-Rincon I, Bueno F, Zambudio G, Poza M. Ventriculopleural shunting with new technology valves. Childs Nerv Syst 2000;16(12):867-871.
5. Megison DP, Benzel EC. Ventriculo-pleural shunting for adult hydrocephalus. Br J Neurosurg 1988;2(4):503-505.
6. Sahn SA. Pleural effusions of extravascular origin. Clin Chest Med 2006;27(2):285-308.
7. Beach C, Manthey DE. Tension hydrothorax due to ventriculopleural shunting. J Emerg Med 1998;16(1):33-36.
8. Mitrouska I, Bouros D. The trans-exudative pleural effusion. Chest 2002;122(5):1503-1505.
9. Haft GF, Mendoza SA, Weinstein SL, Nyunoya T, Smoker W. Use of beta-2-transferrin to diagnose CSF leakage following spinal surgery: a case report. Iowa Orthop J 2004;24:115-118.
10. Sanders DY, Summers R, DeRouen L. Symptomatic pleural collection of cerebrospinal fluid caused by a ventriculopleural shunt. South Med J 1997;90(3):345-346.