thymomas may be cured by such multidisciplinary therapy, including high-dose chemotherapy followed by PBSCT.

REFERENCES

Subarachnoid Pleural Fistula Due to Penetrating Trauma*

Case Report and Review of the Literature

Christian Lloyd, MD; and Steven A. Sahn, MD, FCCP

We describe a case of a 30-year-old man who developed a recurrent pleural effusion after sustaining a gunshot wound to the left side of his chest with subsequent complete paralysis at the T2 level. Subarachnoid-pleural fistulas have rarely been reported as complications of penetrating and blunt trauma, thoracic surgery, as well as spinal surgery. Concomitant injuries may overshadow or complicate the diagnosis of subarachnoid-pleural fistulas. The diagnosis should be considered in any patient with a pleural effusion that is associated with severe neurologic injury, as the fistula rarely heals without surgical intervention and may lead to CNS infection or pneumocephalus.

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Key words: β2-transferrin; CNS infections; CT scanning; hemithorax; laminectomy; myelography; pleural effusion; spinal cord trauma

Abbreviation: CSF = cerebral spinal fluid

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Subarachnoid-pleural fistulas have been reported as complications of penetrating and blunt trauma, and thoracic surgery, as well as spinal surgery. However, it remains a rare complication with, to our knowledge, the present case being only the 30th reported in the literature. Concomitant injuries and dual processes such as hemothorax may overshadow or complicate the diagnosis of subarachnoid-pleural fistula. The diagnosis should be considered in any patient with a pleural effusion associated with severe neurologic injury, as the fistula rarely heals without surgical intervention. Likewise, a persistent subarachnoid-pleural fistula may lead to CNS infection or pneumocephalus.

**Case Report**

A 30-year-old man was transferred to the trauma service at the Medical University of South Carolina Hospital after sustaining a gunshot wound to the left side of his chest. A physical examination revealed complete paralysis and sensory loss at the T2 level. Radiographs showed a bullet lodged at the level of the second thoracic vertebrae. Additional findings included a large left-sided hemothorax. The hemothorax was completely evacuated by tube thoracostomy. On the third hospital day, after drainage decreased to < 70 mL per 24 h with radiographic resolution of the hemothorax, the chest tube was removed. On the third day after the discontinuation of the chest tube, the patient experienced increasing dyspnea and a temperature of 101°F. The findings of a chest examination were consistent with the presence of a left-sided pleural effusion. The peripheral leukocyte count was 19,000 cells/µL, and a chest radiograph revealed a moderate-sized pleural effusion (Fig 1). A second chest tube was inserted, and 1,050 mL fluid was initially collected. At that point, the pulmonary service was consulted.

The pleural fluid was a hemorrhagic exudate. Data from a serial pleural fluid analysis and a cerebral spinal fluid (CSF) analysis, and normal CSF data are shown in Table 1. It was thought that the patient most likely had pneumonia with a parapneumonic effusion, however, the possibility of a subarachnoid-pleural fistula was entertained. To investigate the latter, pleural fluid α1-transferrin levels were measured and the results were negative. Using a cervical approach, a CT myelogram was performed that showed the extravasation of contrast into the left pleural space and the absence of contrast flow past the area of the bullet (Fig 2). It was postulated that with the apparent blockage of the spinal canal and negative pressure of the pleural space with additional chest tube suction, CSF was being drawn into the pleural space through the fistula. The patient underwent a T2 laminectomy with removal of the bullet and patching of the visualized subarachnoid-pleural fistula. He was treated postoperatively with a lumbar drain. The chest tube was placed to water seal, and over the ensuing 72 h the pleural drainage decreased. The chest tube was removed, and a chest radiograph that was obtained prior to hospital discharge confirmed that the effusion had not reaccumulated.

**Discussion**

Although it is a rare complication, subarachnoid-pleural fistulas due to blunt and penetrating trauma represent the
majority of those in the published cases (23 of 30 cases; 77%). In addition, subarachnoid-pleural fistulas have been reported as a complication of transthoracic diskectomy and thoracotomy.\textsuperscript{1,2} The causes of all reported cases and frequencies are shown in Figure 3.

The majority of cases of subarachnoid-pleural fistula are associated with severe spinal cord trauma. A mechanism that results in a traumatic fistula can involve either a missile transversing both the pleural and subarachnoid spaces or a vertebral fracture that tears the dura and the parietal pleura. The fistulous tract usually occurs between the upper thoracic spinal cord and the parietal pleura. In cases of blunt trauma, the disruption occurs due to the extreme extension of the spine, resulting in the tearing of the relatively immobile nerve roots and dura. Likewise, significant chest wall compression may perforate the pleura against the bony prominence of the spine.\textsuperscript{3} Regardless of the cause, the subarachnoid-pleural fistula remains open due to the pressure gradient between the positive pressure in the spinal canal and the negative pressure in the pleural space.

In a review of the literature, Pollack and colleagues\textsuperscript{4} found that 20 of 21 patients (95%) with traumatic subarachnoid-pleural fistulas had complete physiologic cord transection at the time of presentation. Often, there is radiographic confirmation of a concomitant vertebral and

Table 1—Pleural and Cerebrospinal Fluid Analysis in Subarachnoid-Pleural Fistula Related to Trauma\textsuperscript{*}

<table>
<thead>
<tr>
<th>Variables</th>
<th>Thoracentesis</th>
<th>Thoracentesis</th>
<th>Lumbar Puncture</th>
<th>Thoracentesis</th>
<th>Lumbar Puncture</th>
<th>Normal CSF\textsuperscript{15}</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hospital day</td>
<td>9</td>
<td>12</td>
<td>12</td>
<td>16</td>
<td>16</td>
<td>36</td>
</tr>
<tr>
<td>Description of fluid</td>
<td>Bloody</td>
<td>Serosanguinous</td>
<td>Clear</td>
<td>Clear yellow</td>
<td>Clear</td>
<td>Clear</td>
</tr>
<tr>
<td>Nucleated cells</td>
<td>300</td>
<td>3,700</td>
<td>6</td>
<td>588</td>
<td>0</td>
<td>0–5</td>
</tr>
<tr>
<td>Differential cell count, %</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Neutrophils</td>
<td>68</td>
<td>56</td>
<td>60</td>
<td>33</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Lymphocytes</td>
<td>14</td>
<td>7</td>
<td>11</td>
<td>27</td>
<td>63</td>
<td></td>
</tr>
<tr>
<td>Macrophages</td>
<td>2</td>
<td>31</td>
<td>29</td>
<td>14</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mesothelial cells</td>
<td>16</td>
<td>6</td>
<td>7</td>
<td>7</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Eosinophils</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Monocytes</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Protein</td>
<td>1.3 g/dL</td>
<td>1.5 g/dL</td>
<td>16 mg/dL</td>
<td>3.2 g/dL</td>
<td>740 mg/dL</td>
<td>20–50 mg/dL</td>
</tr>
<tr>
<td>PF/S ratio</td>
<td>0.22</td>
<td>0.26</td>
<td>0.55</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>LDH</td>
<td>276 IU/L</td>
<td>451 IU/L</td>
<td>381 IU/L</td>
<td>250 IU/L</td>
<td>40 U/L</td>
<td></td>
</tr>
<tr>
<td>PF/ULLDH ratio</td>
<td>0.93</td>
<td>1.88</td>
<td>1.59</td>
<td>0.1–0.5</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Glucose, mg/dL</td>
<td>121</td>
<td>95</td>
<td>68</td>
<td>56</td>
<td>40–70</td>
<td></td>
</tr>
<tr>
<td>Gram stain and culture</td>
<td>Negative</td>
<td>Negative</td>
<td>Negative</td>
<td>Negative</td>
<td>Negative</td>
<td>Negative</td>
</tr>
<tr>
<td>β2-transferrin</td>
<td>Negative</td>
<td>Negative</td>
<td>Negative</td>
<td>Negative</td>
<td>Positive</td>
<td></td>
</tr>
</tbody>
</table>

*LDH = lactate dehydrogenase; PF = pleural fluid; S = serum protein; ULLDH = upper limits of normal lactate dehydrogenase level.

Figure 2. A CT myelogram showing the extravasation of thecal contrast into the left medial pleural space adjacent to the T2 vertebral body. Medially, the contrast may be extrapleural or loculated within the pleural space. However, contrast is visualized along the posterior-lateral pleura, indicating a thecal-pleural communication.
spinal cord injury. However, children may present with spinal cord injury without radiologic abnormality due to severe extension injuries. A high degree of suspicion is required to establish the diagnosis of subarachnoid-pleural fistula. Often, symptoms related to the effusion such as chest pain, dyspnea, or fever may be minimal or overshadowed by concomitant injuries, and often the diagnosis is delayed. In addition to the almost universal finding of severe spinal cord injury, rare clues to the diagnosis may include pneumocephalus or meningitis. Symptoms that suggest a CSF leak include headache, nausea, and vomiting. The findings of a chest radiograph initially may be normal. Effusions range from small to massive, depending on the size and duration of the effusion. Mediastinal widening also may be seen, and, once aortic injury is excluded, a diagnosis of subarachnoid-medial or subarachnoid-pleural fistula should be considered. Pleural fluid in a subarachnoid-pleural fistula has been described most often as being clear with a low nucleated cell count, having a glucose equivalent to serum, and having a low protein content in the transudative range. However, there may be concomitant pleural processes making pleural fluid analysis problematic, as in the present patient who initially had a hemopneumothorax. Evidence that the hemothorax significantly influenced the pleural fluid analysis may be inferred by the eosinophilia noted in the pleural fluid sample 16 days after the injury (Table 1). Blood in the pleural space is a known cause of an exudative, eosinophilic pleural effusion. The peak pleural fluid eosinophil count tends to occur approximately 7 to 14 days following the entry of blood into the pleural space. During his hospital course, the patient developed fever, chest pain, and worsening oxygenation with a radiographic opacity that was suggestive of pneumonia with parapneumonic effusion. It is postulated that, for these reasons, the pleural fluid analysis was most consistent with an exudate rather than the clear transudate described in the literature. Shannon and colleagues reported a similar case of a 27-year-old stabbing victim who presented with a hemothorax and persistent pleural drainage that was diagnosed as a subarachnoid-pleural fistula. The pleural fluid continued to have a high protein concentration 2 months after the closure of the pleural fistula. As in our case, electrophoresis for β₂-transferrin failed to identify CSF in the pleural fluid. β₂-transferrin has been widely used to identify CSF leakage following head trauma, as it is found only in CSF and inner ear perilymph fluid. The test is both sensitive (100%) and specific (95%) for CSF leaks due to head trauma. β₂-transferrin never has been studied in the setting of a subarachnoid-pleural fistula, and we postulate that concomitant pleural processes may lead to a false-negative result.

Definitive identification of a fistula is confirmed radiographically. Over the past 40 years, the quality of imaging modalities has improved. Initially, the direct infusion of dye into the thecal sac with subsequent aspiration of pleural fluid was used to establish the diagnosis. Myelography added the advantage of being able to visualize the anatomic defect. However, there are reports of false-negative results using myelography alone due to slow or intermittent leaks. CT scanning performed concomitantly with myelography adds to the delineation of the anatomic defect. In addition, the presence of contrast in the pleural space after myelography confirms the diagnosis in patients with slow CSF leaks. Isotope myelography, a sensitive test, also has been used to confirm the diagnosis. Any isotope detection in the pleural space after injection into the spinal canal confirms the diagnosis, although it does not provide anatomic information.

There is no consensus for the management of subarachnoid-pleural fistulas. As spontaneous resolution is rare, virtually all patients require closed-tube drainage, and most require surgical correction of the fistula. Of the 19 posttraumatic subarachnoid-pleural fistulas reported by Pollack and colleagues, 13 (68%) were treated definitively with laminectomy or thoracotomy, while 3 (16%) responded to chest tube drainage. Two of 19 subarachnoid-pleural fistulas (11%) were treated with thoracotomy, while 1 was treated with “conservative measures.” In 9 of 11 patients (82%) in whom a laminectomy was performed, the closure of the fistula was successful. The appropriate timing of surgical intervention is unknown. Some surgeons advocate chest tube drainage for up to 2 weeks before surgical intervention, while others recommend early intervention, noting the low rate of spontaneous closure.

References

Cardiac Decortication (Epicardiectomy) for Occult Constrictive Cardiac Physiology After Left Extrapleural Pneumonectomy*

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Constrictive cardiac physiology typically does not occur in the absence of parietal pericardium. However, we report eight patients who, after left extrapleural pneumonectomy and removal of the parietal pericardium for malignancy, presented with dyspnea, jugular venous distension, and peripheral or generalized edema unresponsive to diuretics. Cardiac decortication (epicardiectomy) was performed whereby a thickened peel encasing the heart was surgically excised, resulting in vigorous contraction and expansion of the heart. In one patient, decortication occurred early after pneumonectomy and was incomplete. Acute signs of inflammation were present, and recurrence necessitated repeat decortication. When patients present with dyspnea, hepatojugular reflux, and peripheral edema refractory to diuretics, constrictive cardiac physiology should be considered in the differential diagnosis, even in the absence of parietal pericardium.

(CHEST 2002; 122:2256–2259)

Key words: cardiac decortication; epicardiectomy; pneumonectomy

Abbreviations: EPP = extrapleural pneumonectomy; POD = postoperative day; RV = right ventricular

Extrapleural pneumonectomy of the left lung includes removal of the lung, parietal pleura, parietal pericardium, and diaphragm.1 Therefore, constrictive cardiac physiology due to pericarditis would not seem possible postoperatively. We report eight patients who nonetheless acquired occult constrictive cardiac physiology due to epicardial constriction, despite left extrapleural pneumonectomy (EPP) for underlying cancer. In all cases, fibrous material grew around and encased the heart. These patients required reoperation for epicardial decortication to alleviate dyspnea and peripheral edema refractory to diuretics.

Materials and Methods

We undertook a retrospective chart review of the preoperative presentations, clinical evaluations that revealed constrictive physiology, operative procedures, and patient outcomes.

Results

In the period from February 1997 to January 2000, 133 patients underwent EPP. Seven patients (5%) acquired constrictive cardiac physiology after left EPP for malignant mesothelioma, and one patient acquired constrictive cardiac physiology after left intrapericardial pneumonectomy for non-small cell lung carcinoma. The mesothelioma was epithelial in six patients and epithelial/sarcomatoid in one patient. The parietal pericardium was entirely excised in seven patients and partially excised in one patient. Pericardial and/or diaphragm reconstruction was accomplished using polytetrafluoroethylene (Gore-Tex; W.L. Gore & Associates; Flagstaff, AZ). Adjunct therapy included preoperative chemotherapy and radiation in one patient and intraoperative heated chemotherapy in four patients. No talc was used in any patients.

After a median interval of 3.3 months (range, 1.6 to 18.9 months), patients required reoperation for recurrent pericardial constriction.