

# Treatment of the subarachnoid–pleural fistula

## Case report

SERDAR OZGEN, M.D., BURAK O. BORAN, M.D., ILHAN ELMACI, M.D., UGUR TURE, M.D., AND M. NECMETTIN PAMIR, M.D.

*Marmara University, Department Of Neurosurgery, Istanbul-Turkey*

Subarachnoid–pleural fistula is a rare type of cerebrospinal fluid (CSF) fistula, and there are only several cases reported in the literature. The authors describe a 65-year-old male patient in whom a diagnosis of T7–8 disc herniation had been made. He underwent surgery via a right lateral extracavitary approach. Postoperatively he developed progressive respiratory distress and headache. A chest x-ray film revealed a pleural effusion, and computerized tomography (CT) myelography demonstrated a subarachnoid–pleural fistula at the level at which the herniated disc had been removed. The patient had been managed via a CSF drainage system and a chest tube. He was discharged after relief of symptoms was attained. Subarachnoid–pleural fistulas can be secondary to traumatic injury and surgery, or they can be spontaneous. Patients present with rapidly filling pleural effusion and headache. A diagnosis can be established using CT myelography or myeloscintigraphy. Treatment is conservative, with the placement of a chest tube and insertion of a CSF drainage catheter, and surgical repair should be considered only if the conservative therapy fails.

**KEY WORDS** • subarachnoid–pleural fistula • thoracic spine • extracavitary approach

Subarachnoid–pleural fistula is a rare type of CSF fistula. There have only been 54 cases reported in the literature.<sup>1–6,8–30</sup> The fistula may develop secondary to traumatic injury<sup>1–3,5–8,11,14,15,17,20–24,26,29,30,32–42,44,47–52,</sup> or surgery.<sup>4,9,10,12,13,16,18,19,25,27,28,34,43,45,53,54</sup> Although it is rare, because of its severe clinical picture, diagnosis and treatment are of vital importance. A case of subarachnoid–pleural fistula is presented, and we review the literature, concentrating on the clinical presentation, diagnostic modalities, and treatment strategies.

### CASE REPORT

**Presentation.** This 65-year-old male presented with a long history of lumbar and bilateral radicular leg pain. He had previously undergone a laminectomy for the treatment of T10–11 disc disease.

**Examination.** Physical examination demonstrated bilateral increased deep tendon reflexes and hypesthesia beneath the level of T-10. His MR images revealed a thoracic disc herniation narrowing the spinal canal at the level of T7–8.

**Operaton.** He underwent surgery via a right lateral extracavitary approach. The right-sided T-8 nerve root had

been sectioned and root sleeve tied. The intervertebral disc was calcified and adherent to dura anteriorly. Following the T7–8 discectomy a CSF leak was observed, but after the intervertebral space had been packed with Curaspon, an absorbable gelatin sponge (Clinimed Holding Ijweg 91, 1161 ET Zwanenburg, The Netherlands) the leakage ceased. Positive abdominal pressure has been applied by forced ventilation, and no leakage was detected. Additionally a 1 to 2–cm tear in the pleura was noticed but not repaired.

**Postoperative Course.** On the 3rd postoperative day, the patient developed progressive respiratory distress and headache. His chest x-ray film revealed a pleural effusion. The effusion was drained via a tap. The fluid was slightly hemorrhagic, and the biochemical profile was as follows: glucose 59 mg/dl; pleural fluid/serum protein ratio 0.17; and pleural fluid/serum LDH ratio 0.34 (which indicated that the fluid was a transudate). Two days later, the effusion rapidly filled again. A subarachnoid–pleural fistula was suspected. A lumbar external drainage catheter was inserted at L4–5, and the position of the catheter was checked by administration of a contrast material under fluoroscopy. However only few milliliters of CSF could be drained. Computerized tomography myelography revealed the fistula at the surgical level (Fig. 1).

**Second Operation.** The patient underwent surgery, and the pleura was repaired using muscle flap. Cerebrospinal fluid was leaking from the intervertebral space, but be-

Abbreviations used in this paper: CSF = cerebrospinal fluid; CT = computerized tomography; LDH = lactate dehydrogenase; MR = magnetic resonance.

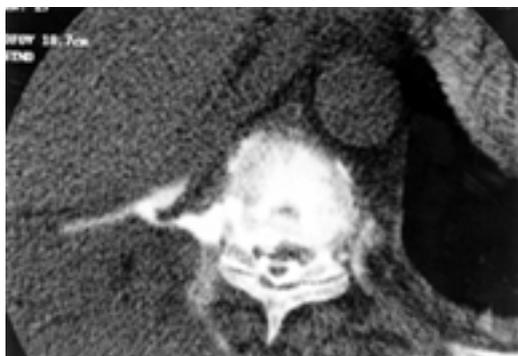


Fig. 1. Computerized tomography myelography demonstrating the subarachnoid–pleural fistula at the level where surgery had previously been performed.



Fig. 2. Chest x-ray film revealing the massive pleural effusion 4 days after the initial surgical repair of the pleura.

cause the dural defect could not be visualized, the space was sealed with fibrin glue and packed with Curaspon. A CSF drainage catheter was inserted under direct vision, at the level of T6–7. However, 4 days later, the effusion filled again, and this time it was massive (Fig. 2). A chest tube was applied. The CSF drainage system was kept in place for 7 days and the chest tube for 12 days.

**Second Postoperative Course.** The patient received parenteral vancomycin and imipenem treatment. He experienced no additional problems. He was followed for 2 weeks in the ward and was discharged after his symptoms were relieved.

## DISCUSSION

Subarachnoid–pleural fistula is a rare type of CSF fistula. The first case was reported in 1959 by Milloy, et al.<sup>33</sup> A search of Medline records revealed that since that first publication 54 cases have been reported. Thirty-seven were secondary to traumatic injury,<sup>1–3,5–8,11,14,15,17,20–24,26,29,30,32–42,44,47–52,55</sup> and 15 to surgery.<sup>4,9,10,12,13,16,19,25,27,28,34,43,45,53,54</sup>

There was one case in which the fistula developed secondary to thoracoscopy,<sup>18</sup> and one spontaneous case secondary to rupture of an intrathoracic meningocele.<sup>46</sup> Six of the surgery-related fistulas were complications of the lateral extracavitary approach to the thoracic spine. The preoperative diagnoses in these patients were as follows: three cases of thoracic disc disease,<sup>13,25,34</sup> two cases of schwannoma,<sup>12,43</sup> and one case of neurofibroma.<sup>45</sup>

### Clinical Presentation

Patients typically present with respiratory stress, and chest x-ray films reveal the pleural effusion. If a spinal tap is performed it reveals the characteristics of a transudate. Monla-Hassan, et al.,<sup>33</sup> have postulated that the biochemical evaluation of a pleural effusion that was drained represented CSF. In our opinion, it is impossible to distinguish between CSF and any other transudational pleural effusion. According to the criteria proposed by Light, et al.,<sup>31</sup> a transudate is described as having pleural fluid/serum protein ratio less than 0.5, a pleural fluid/serum/LDH ratio less than 0.6, and pleural fluid LDH less than two thirds of the upper limit of normal serum value; therefore CSF and any other pleural transudate cannot be distinguished biochemically.

In several reported cases, headache accompanied pleural effusion.<sup>4,10,12,18,21</sup> The headache is caused by two different mechanisms. If the parietal pleura is defective but the visceral pleura is intact, the pressure of the pleural space ranges from  $-5$  to  $-7.5$  cm H<sub>2</sub>O. On the other hand, pressure of CSF is 10 to 15 cm H<sub>2</sub>O. This leads to a suction effect, and CSF diverts into the pleural space, which decreases CSF pressure and produces headache.<sup>12</sup> On the other hand, if the visceral pleura is defective, the pressure in the pleural space, overcomes the CSF pressure during expiration and air leaks into the subarachnoidal space which produces pneumocephalus.<sup>4,10,21</sup> Boyev, et al.,<sup>4</sup> have presented a case in which the pneumocephalus was so prominent that the presenting clinical picture consisted of gait disturbance and confusion. Air within the ventricles has been demonstrated by CT scanning in few cases.<sup>4,18,21</sup>

### Diagnostic Modalities

Diagnostic procedures for the demonstration of the subarachnoid–pleural fistulas include CT myelography, myeloscintigraphy, and MR imaging. Computerized tomography myelography is easy and very effective in demonstrating the fistula. Peter and Rode<sup>40</sup> have postulated that MR imaging is superior to both the aforementioned modalities, and it is especially valuable in demonstrating the associated spinal cord damage in traumatically injured patients, but analysis of the literature indicates that it is not a widely accepted diagnostic tool in these cases.

### Treatment Strategies

Choosing the treatment strategy is the most challenging aspect of the case. Several authors<sup>6,11,13,14,53</sup> have postulated that conservative therapy is suitable in most of the patients. However, other authors<sup>15,19,26,48</sup> have argued that surgical repair of the fistula is essential and that early intervention in these cases is rewarding. There are also other suggested treatment options. Díaz, et al.,<sup>12</sup> have reported placing a pleuroperitoneal shunt that was not successful. Katz, et al.,<sup>27</sup> have reported that they successfully treated a case by placing lumboperitoneal shunt to drain the fistula.

External lumbar drainage is generally an effective treatment technique in the management of patients with any type of CSF fistula. In our case a lumbar catheter was

## Subarachnoid–pleural fistula

inserted initially at L4–5, but effective CSF drainage could not be established because the CSF was sucked up by the pleural space due to the pressure difference. During the surgical repair of the pleura, a CSF drainage system was applied at T6–7, and it was supposed to work because CSF circulates downwards posterior to the spinal cord and then upward anterior to the spinal cord. Even when the catheter was introduced surgically from above the level of the fistula, the CSF could not be drained. Therefore, CSF drainage systems should be used in conjunction with a chest tube. Takenouchi, et al.,<sup>53</sup> have stated that surgical repair cannot be successful if a chest tube is not inserted postoperatively. Another question is when to remove the chest tube and the drainage catheter. Seven days of CSF drainage is adequate for the closure of the dural defect, but the chest tube should be kept in place for several additional days to protect the recently closed dural defect from pressure differences. Dickman, et al.,<sup>13</sup> also stated that the CSF drainage system should be removed first and the chest tube should be left in place for a few more days.

In conclusion, although the subarachnoid–pleural fistula is a rare type of CSF fistula, its potential diagnosis should always be kept in mind. It is best diagnosed using CT myelography or myeloscintigraphy. Conservative therapy, which consists of placement of a chest tube and a CSF drainage system, should be the initial treatment strategy in every patient, and surgical repair of the fistula should be considered only if the conservative treatment fails.

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Address reprint requests to: Serdar Ozgen, M.D., PK: 4 81082, Goztepe, Istanbul, Turkey. email: omurga@turk.net.