Unilateral massive transudative pleural effusion due to a duropleural fistula

Dura-plevral fistüle bağlı tek taraflı masif transüdatif plevral efüzyon

İpek Özmen,¹ Emine Aksoy,¹ Nilüfer Kongar,¹ Güliz Ataç,¹ Tülin Sevim,¹ Kadriye Terzioğlu,² Erhan Çelikoğlu,³ Kemal Tahaoğlu⁴

Institution where the research was done: Department of Chest Diseases, Chest Diseases and Thoracic Surgery Training and Research Hospital, İstanbul, Turkey

Author Affiliations:

¹Department of Chest Diseases, Chest Diseases and Thoracic Surgery Training and Research Hospital, İstanbul, Turkey ²Department of Chest Diseases, Mardin State Hospital, Mardin, Turkey

³Department of Neurosurgery, Dr. Lütfi Kırdar Kartal Training and Research Hospital, İstanbul, Turkey

⁴Department of Chest Diseases, Anadolu Medical Center, İstanbul, Turkey

Twenty three-year-old man had thoracal spinal cord surgery for gunshot injury three months ago. He had right hemiplegia and paraplegia for the past two months. He was admitted with poor performance status and progressive, severe dyspnea. Chest X-ray showed massive pleural effusion on the right side. Thoracentesis showed transudative pleural fluid. Duropleural fistula was considered due to the history of spinal injury and surgery. The presence of cerebrospinal fluid in the pleural fluid was demonstrated by beta-2 transferrin test. After the demonstration of the presence of duropleural fistula on intrathecal contrast-enhanced computed tomography scan surgical management of the duropleural fistula was successfully repaired by duroplasty. This case was presented as a complication of spinal cord injury and neurosurgery duropleural fistula is a very rare reason of massive transudative pleural effusions and the use of the cerebrospinal fluid by beta-2 transferrin in pleural effusion is reported very rare in the literature.

Keywords: Beta-2 transferrin; duropleural fistula; transudative effusion.

Pleural effusion is a common clinical presentation in pulmonary medicine. Exudative effusions are generally seen as manifestations stemming from pulmonary issues, whereas transudative effusions usually occur because of nonpulmonary or systemic Yirmi üç yaşında erkek hasta üç ay önce ateşli silah yaralanması nedeni ile torakal spinal cerrahi geçirmiş idi. Hastanın son iki aydır sağ hemiplejisi ve paraplejisi var idi. Hasta kötü performans statüsü ve ilerleyici, siddetli nefes darlığı ile başvurdu. Akciğer grafisinde sağ tarafta masif plevral efüzyon izlenmekte idi. Torasentez ile transüdatif plevral sıvı saptandı. Spinal yaralanma ve cerrahi öyküsü nedeni ile dura-plevral fistül düşünüldü. Plevra sıvısında beyin omurilik sıvısının varlığı beta-2 transferrin testi ile gösterildi. Intratekal kontrastlı bilgisayarlı tomografi taramasında dura plevral fistül varlığı gösterildikten sonra fistül, duraplasti işlemi ile başarılı bir şekilde kapatıldı. Bu olgu, spinal cerrahi ve spinal yaralanmalar sonrası bir komplikasyon olarak oluşabilen dura-plevral fistül transüdatif plevral efüzyonun nadir bir nedeni olması ve beyin omurilik sıvısının plevra sıvısında varlığının gösterilmesinde beta-2 transferrin kullanımı literatürde az sayıda bildirilmesi nedeni ile sunuldu.

Anahtar sözcükler: Beta-2 transferrin; dura-plevral fistül; transüdatif efüzyon.

reasons. Herein, we present the case of a male patient with transudative pleural effusion caused by an extremely rare duropleural fistula (DPF). He presented with progressive dyspnea and was diagnosed using pleural beta-2 transferrin.



Available online at www.tgkdc.dergisi.org doi: 10.5606/tgkdc.dergisi.2014.7605 QR (Quick Response) Code Received: August 28, 2012 Accepted: November 05, 2012

Correspondence: İpek Özmen, M.D. Süreyyapaşa Göğüs Hastalıkları ve Göğüs Cerrahisi Eğitim ve Araştırma Hastanesi, Göğüs Hastalıkları Kliniği, 34854 Maltepe, İstanbul, Turkey.

Tel: +90 216 - 421 42 00 e-mail: ipekozmen2011@gmail.com

CASE REPORT

A 23-year-old man underwent spinal cord surgery at the T_3 - T_4 level because of a gunshot injury three months prior to coming to our facility. In addition, for the last two months, he had been receiving physical therapy at a rehabilitation hospital because of right hemiplegia and paraplegia. After also suffering from progressive severe dyspnea for a week, he was then admitted to our clinic.

A physical examination revealed that he had poor performance and severe dyspnea. His arterial blood pressure was 90/60 mmHg, and he had pulse rate of 130 minute, a respiratory rate of 26 minute, an oxygen saturation (OS) rate of 88%, and a body temperature of 36.6 °C.

A chest examination found evidence of massive pleural effusion on the right side, which was confirmed by a chest radiograph. In addition, it was causing a mediastinal shift to the left (Figure 1). The patient also had right hemiplegia and paraplegia, but there was no edema on the extremities.

The results of the serum laboratory examination of the hematological and biochemical parameters were within normal limits. He had a white blood cell (WBC) count of 6.611 mm³ and a red blood cell (RBC) count of 5.00 mm³. Furthermore, his hemoglobin (Hgb) was 14.7 g/dL, and he had a hematocrit (Hct) level of 42.8%. Additionally, his platelet (PLT) count was 342.000 μ l, and his erythrocyte sedimentation rate (ESR) was 15 mm hour. A thoracentesis showed a very light clear, waterlike fluid made up of 84 mg/dL glucose, 2.6 g/dL total protein, 1.7 g/dL albumin, and 59 IU/L lactate dehydrogenase (LDH). Furthermore, the serum laboratory results revealed a glucose level of 82 mg/dL, a total protein level of 8.9 g/dL, an albumin level of 4 g/dL, and an LDH level of 176 IU/L. A pleural fluid cytological study was unremarkable.

The common causes of transudative effusion were excluded after the clinical evaluation and laboratory investigations, and we hypothesized that the DPF was due to his recent spinal cord injury and surgical history. Because of the patient's severe dyspnea and poor performance, we could not perform a computed tomography (CT) myelogram, or magnetic resonance imaging (MRI). Furthermore, the presence of cerebrospinal fluid (CSF) was demonstrated by determining the levels of beta-2 (β 2) transferrin when the analysis of the pleural effusion.

Since the patient was suffering from dyspnea with massive pleural effusion, a chest tube was inserted, and ceftriaxone was administered as a prophylactic for meningitis. Afterwards, the patient was referred to the neurosurgery clinic where a pseudomeningocele and the DPF at the T_3 - T_4 level were detected via intrathecal contrast-enhanced CT (Figure 2). Repair of the DPF was then successfully performed by duroplasty using muscle, fascia, and tissue glue.

DISCUSSION

The disrupture of the dural membrane and parietal pleura by either a spinal cord injury or neurosurgery result in a leak between the subarachnoid and



Figure 1. Chest X-ray showing the massive pleural effusion on the right side that was causing a mediastinal shift to the left.



Figure 2. Intrathecal contrast-enhanced computed tomography showing the subarachnoid pleural fistula. The white arrow indicates the leakage of the contrast from the subarachnoid space into the pleural space.

pleural spaces. The negative pressure in the pleural space and positive pressure in the subarachnoid space allow for the accumulation of CSF in the pleural space,^[1] which usually is clear and has low protein levels (<1 g/dL), and low nucleated cells in the nontraumatic DPF. However, the diagnosis may be delayed in cases involving a traumatic hemothorax.^[1-3]

Symptoms may vary according to the amount of CSF that collects in the pleural space, with postural headaches, nausea, vomiting, dyspnea, and chest pain being common. Symptomatic large pleural effusions due to the DPF are very rare.^[2-4] Our patient had hemiplegia and paraplegia, and his immobility might be the reason why he stayed asymptomatic until the accumulation of the massive pleural effusion caused the mediastinal shift.

Beta-2 transferrin is a protein produced by neuraminidase activity in the brain that is found in CSF, ocular fluids, and inner ear perilymph. It is also an important marker of CSF leakage.^[5-7] This type of transferrin is widely used by otolaryngologists postoperatively to determine CSF rhinorrhea, and it provides a rapid, noninvasive way of detecting this fluid. The sensitivity of beta-2 transferrin ranges from 94-100%, and its specificity is between 98 and 100%.^[8] A CT myelogram or MRI can be used to determine the location of DPFs,^[3,4,7] but in our case they could not be performed because of severe dyspnea and poor performance along with the possibility that some of the metallic particles from his gunshot wound might not have been removed.

There are only a few reports in the literature that have focused on the diagnostic use of beta-2 transferrin for the diagnosis of DPFs.^[1,9] Huggins and Sahn^[1] conducted the first study of this kind which featured an 81-year-old man who developed chronic transudative pleural effusion after lumbar disc surgery had been performed two years before. In another study by Lloyd et al.,^[10] a patient with a subarachnoid pleural fistula due to gunshot trauma initially presented with a hemopneumothorax. However, the pleural fluid beta-2 transferrin test was negative, and a CT myelogram then diagnosed a subarachnoid pleural fistula.

The case was presented, as a complication of spinal cord injury and neurosurgery duro-pleural fistula is

a very rare reason of massive transudative pleural effusions and determination of the cerebrospinal fluid by beta-2-transferrin in pleural effusion is reported very rare in literature. Besides that further investigations may needed in this subject.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

- 1. Huggins JT, Sahn SA. Duro-pleural fistula diagnosed by beta2-transferrin. Respiration 2003;70:423-5.
- Monla-Hassan J, Eichenhorn M, Spickler E, Talati S, Nockels R, Hyzy R. Duropleural fistula manifested as a large pleural transudate: an unusual complication of transthoracic diskectomy. Chest 1998;114:1786-9.
- 3. D'Souza R, Doshi A, Bhojraj S, Shetty P, Udwadia Z. Massive pleural effusion as the presenting feature of a subarachnoid-pleural fistula. Respiration 2002;69:96-9.
- 4. Ozgen S, Boran BO, Elmaci I, Ture U, Pamir MN. Treatment of the subarachnoid-pleural fistula. Case report. Neurosurg Focus 2000;9:ecp1.
- Meurman OH, Irjala K, Suonpää J, Laurent B. A new method for the identification of cerebrospinal fluid leakage. Acta Otolaryngol 1979;87:366-9.
- 6. Skedros DG, Cass SP, Hirsch BE, Kelly RH. Beta-2 transferrin assay in clinical management of cerebral spinal fluid and perilymphatic fluid leaks. J Otolaryngol 1993;22:341-4.
- 7. Papadea C, Schlosser RJ. Rapid method for beta2transferrin in cerebrospinal fluid leakage using an automated immunofixation electrophoresis system. Clin Chem 2005;51:464-70.
- Warnecke A, Averbeck T, Wurster U, Harmening M, Lenarz T, Stöver T. Diagnostic relevance of beta2-transferrin for the detection of cerebrospinal fluid fistulas. Arch Otolaryngol Head Neck Surg 2004;130:1178-84.
- 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;24:115-8.
- 10. Lloyd C, Sahn SA. Subarachnoid pleural fistula due to penetrating trauma: case report and review of the literature. Chest 2002;122:2252-6.