

## Persistent Chylothorax in Behcet's Disease Treated with a Blood Patch

Hannah Maayan MD<sup>1</sup>, Gabriel Izbicki MD<sup>2</sup>, Judith Heyd MD<sup>1</sup>, Ruth Cyter MD<sup>3</sup>, Shuli Silberman MD<sup>4</sup> and Maher Deeb MD<sup>4</sup>

Departments of <sup>1</sup>Ocology-Hematology, <sup>2</sup>Pulmonology, <sup>3</sup>Radiology and <sup>4</sup>Cardiothoracic Surgery, Shaare Zedek Medical Center, and Hebrew University Medical School, Jerusalem, Israel

**Key words:** Behcet's disease, chylothorax, blood patch, venous thrombosis, chest tube

*IMAJ 2008;10:588–589*

Chylothorax is a rare complication of Behcet's disease and many treatment options have been described. We report a case treated successfully with a blood patch introduced into the chest via an intercostal chest drain.

### Patient Description

A 21 year old male was admitted in December 2003 to our hospital because of a severe sore throat and an immobile neck. His medical history was remarkable for occasional oral and genital ulcers. On admission he was in severe distress, with a low grade fever of 37.2–38°C. Examination of the neck and pharynx revealed trismus, enlarged cervical lymph nodes and tonsils with no exudates. Marked venous collaterals were noted on his arms and chest. Routine laboratory work was within normal limits with increased inflammatory markers. Computed tomography scan of the neck showed swollen tonsils with retropharyngeal edema and thrombi in both jugular veins. The thrombus on the left side filled the jugular vein from the jugular foramen to the entrance of the superior vena cava.

Antibiotic and anticoagulation treatment was initiated, but after 2 weeks there was no improvement. The patients' history of recurrent oral and genital ulcers was reconsidered and a positive pathergy test supported the diagnosis of Behcet syndrome. Treatment with oral steroids was added, leading to rapid amelioration of all symptoms.

Three days after discharge he was readmitted with shortness of breath and a productive cough. A CT scan of the neck and chest [Figure] revealed a massive left

pleural effusion with passive atelectasis of the left lung. Two liters of chylous fluid were drained. His treatment was modified by adding a small dose of methotrexate (7.5 mg/week) and encouraging a diet of middle-chain fatty acids. Despite this conservative treatment, chylothorax recurred and weekly pleural taps were inserted.

Surgical intervention was considered but deemed risky due to the extensive venous collaterals on the chest wall and increased risk of excessive bleeding. Instead, a 12 French intercostal chest tube was inserted into the pleural cavity. After complete drainage of the pleural cavity, 50 ml of autologous blood were injected via the chest tube. The tube was clamped for 3 hours and connected to suction. The procedure was repeated in the next 2 days, after which chyle discharge stopped and the chest tube was removed. Follow-up of 12 months showed no reaccumulation of chyle.

### Comment

Behcet disease is a systemic disease primarily affecting the eye and genital mucosa. Diagnosis is based on established criteria proposed by the International Study Group for Behcet Disease in 1990. These include the presence of recurrent

oral ulcers plus at least two of the following – recurrent genital ulceration, eye lesions, skin lesions, or a positive pathergy test [1]. Vascular complications are more common in male patients and include thromboembolism, arterial aneurysms and arterial occlusions [2]. Venous complications are more common and range from superficial thrombophlebitis to deep vein thrombosis of large veins including superior vena cava syndrome [2]. Treatment consists of steroids and occasionally immunosuppression. Anticoagulation is added in cases of thromboembolism or arterial occlusion. Chylothorax is usually the result of massive venous thrombi obstructing the drainage of the thoracic duct [3].

Conservative treatment of chylothorax as a complication of Behcet disease has



CT scan of the chest showing a left pleural effusion

been reported to be effective, with immunosuppression anticoagulation and a medium-chain fatty acid diet [3]. If there is no improvement despite conservative treatment, a more invasive approach may be necessary. Oz et al. [3] report a case in which a pericardial window was created by video-assisted thoracic surgery to drain pericardial fluid and to perform pleural abrasion and drainage of pleural fluid.

Blood outside its own environment is a strong irritant; its injection is painless and causes no side effects. In a report of 25 patients with recurrent or persistent pneumothorax, one to three injections of 50 ml of blood were given to induce pleurodesis. During a follow-up of 2–11 years there were only four recurrences (16%) [4]. In a prospective study of 167 patients who underwent drainage for spontaneous pneu-

mothorax, 32 with autologous blood patch pleurodesis and the others with standard drainage. The authors concluded that the duration of the air leak was significantly shorter in patients who underwent this procedure and there were few complications [5].

Due to the high risk of surgical intervention in this patient, we elected to seal the pleural space with injection of autologous blood. To the best of our knowledge this is the first report of an autologous blood injection as a successful pleurodesis agent in the treatment of recurrent chylothorax.

## References

1. International Study Group for Behcet's Disease Criteria for diagnosis of Behcet's disease. *Lancet* 1990;335:1078–80.

2. Sharir M, Isakov I. Behcet's disease – current status. *IMAJ* 2002; 4(Suppl):946–8.
3. Oz N, Sarper A, Erdogan A, Demircan A, Isin E. Video-assisted thoracic surgery for the management of pleural and pericardial effusion in Behcet syndrome. *Tex Heart Inst J* 2000;27:304–6.
4. Robinson C. Autologous blood for pleurodesis in recurrent and chronic spontaneous pneumothorax. *Can J Surg* 1987;30:428–9.
5. Cagirici U, Sahin B, Cakan A, Kayabas H, Buduneli T. Autologous blood patch pleurodesis in spontaneous pneumothorax with persistent air leak. *Scand Cardiovasc J* 1998;32:75–8.

**Correspondence:** Dr. M. Deeb, Dept. of Cardiothoracic Surgery, Shaare Zedek Medical Center, Jerusalem 91031, Israel.

Phone: (972-2) 655-5918; Fax: (972-2) 652-8934  
email: deeb@szmc.org.il

*Rather than love, than money, than fame, give me truth*

Henry David Thoreau (1817-1862), American naturalist and author

*Invention requires an excited mind; execution, a calm one*

Johann Peter Eckermann (1792-1854), German poet and author, best known for his association with the poet Johann Wolfgang von Goethe