20°C (Table 1). Then the lung block was placed in a bag filled with 1 L of cold Perfadex (Vitrolife, Göteborg, Sweden) and stored in cold saline until transplantation. The cold ischemic time after EVLP was 3 hours for the right and 5 hours for the left lung. Total operation time was 5 hours and 30 minutes. The recipient did not receive any blood transfusion. The patient was extubated 6 hours after the transplantation. Intensive care unit stay was 3 days. Primary graft dysfunction score at postoperative time 48 hours and 72 hours was 0. The last chest tube was removed at postoperative day 6. The patient was discharged from the hospital at postoperative day 21. The FEV 1 at discharge was 2.8 l (64%). The patient is alive and follow-up time is 6 weeks posttransplant.

Comment

We report a successful lung transplantation after reconditioning and treatment with urokinase during EVLP. Perfusion of grafts from non-heart-beating donors (NHBD) shows typically high resistance and inadequate microperfusion suggesting the presence of thrombi and fibrin deposition in the microcirculation [4]. Plasminogen activators are such agents utilized for this purpose. Adding a plasminogen activator, urokinase, into the perfusion solution during ex vivo perfusion and evaluation after 3 hours of warm ischemia resulted in improved graft function and significant reduction of pulmonary vascular resistance (PVR) [4]. This effect of urokinase has been attributed to reconditioning of the graft by dissolving microthrombi [4]. Improvements in pH, PO2, and partial pressure of carbon dioxide with EVLP all may have led to reductions in PVR in addition to the effect of urokinase, which has contributed to the dissolving of microthrombi. In our case, the donor had initially central pulmonary emboli, which were treated by an intravenous thrombolytic agent. Thorax CT scan, which was performed for donor evaluation, showed lyses of central emboli but persistent peripheral arterial lung emboli could not be ruled out. For this reason, we added urokinase into the perfusion solution during EVLP. As seen in Table 1, PVR dropped from 4.9 to 3.7 Wood Units, as well as excellent lung compliance. Lung compliance during retrieval was 42 mL/cm H2O, improved to 74 mL/H2O, and was stable during EVLP. Oxygenation remained stable. Reduction of PVR is a very important indicator for dissolving microemboli [4]. In the early postoperative period, we did not observe any problems such as allergic or toxic effects, or bleeding-related complications to urokinase.

Primary graft dysfunction score at 48 and 72 hours was 0, showing an excellent graft function. In a research project the use of EVLP to evaluate a donor lung graft with acute pulmonary embolism has been recently reported [5]. The authors observed decreased PVR, increased lung compliance, and oxygenation during EVLP. In conclusion, our case report may encourage other groups which utilize NHBD category 1 or 2 donors (other than brain death donors with pulmonary embolism) to add urokinase in the perfusion solution during EVLP.

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References


Pneumothorax as the Initial Manifestation of Idiopathic Hypereosinophilic Syndrome

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We report a case of hypereosinophilic syndrome in a 47-year-old man who had acute pneumothorax as the initial presentation. Peripheral blood eosinophil count increased continuously over a period of 1 month and was associated with pulmonary changes and appearance of skin lesions on the right chest wall. Idiopathic hypereosinophilic syndrome was confirmed by bone marrow aspiration biopsy and skin lesion biopsy after exclusion of all possible secondary etiologies. The clinical status and chest radiographs showed marked improvement after treatment with corticosteroids.


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Hypereosinophilic syndromes (HES) are a rare and heterogeneous group of disorders characterized by persistent marked peripheral or tissue eosinophilia, or both, and eosinophil-mediated organ dysfunction [1]. Lung is frequently involved in HES, and the most common radiologic manifestations include nodules, ground-glass opacities, interlobular septal thickening, and pleural effusion [2]. To our knowledge, however, there is no reported case of HES presenting with pneumothorax. Here, we report the case of a middle-aged man with hypereosinophilia-associated pneumothorax that responded satisfactorily to corticosteroid therapy.

A 47-year-old manual worker who presented with 3-day history of fever, cough, chest pain, chest distress, and shortness of breath was admitted to a local hospital. He had been healthy before admission, with no notable history of respiratory or allergic disease. On admission, routine blood tests showed eosinophil count of 0.82 \( \times 10^9/L \) (normal range, 0.05 to 0.45 \( \times 10^9/L \)), and chest plain X-ray film showed left pneumothorax (Fig 1A). A focal opacity in the lower lobe of the left lung and bilateral pleural effusions were detected on computed tomography scan (Fig 1B). On day 4, the patient had a sudden right pneumothorax. Bilateral pneumothorax was relieved after closed thoracic drainage, but peripheral blood eosinophil count increased to 2.10 \( \times 10^9/L \) in the following days. Computed tomography scan on day 13 demonstrated a newfound opacity in the upper lobe of the right lung.

The patient was transferred to our hospital on day 19, presenting with chest pain, shortness of breath, and a low fever. He denied any history of drug abuse, insect bite, or travel history in the past 3 months. Peripheral blood eosinophil count was 5.86 \( \times 10^9/L \) on admission, and increased to 7.75 \( \times 10^9/L \) on day 27. On day 30, migratory opacities were still evident in both lungs on the computed tomography scan, and several small erythematous nodules emerged on the skin of the right chest wall. A biopsy from the skin lesions showed infiltration of a large number of eosinophils into the fibroadipose tissue (Figs 2B, 2C). The diagnosis of HES was made after the exclusion of secondary etiologies of eosinophilia, such as allergy, parasitic infection, and connective tissue disorders. Furthermore, bone marrow aspiration biopsy showed active proliferation of marrow cells, with increased percentage of eosinophils (Fig 2C), and there was no clonal marker of myeloid or lymphoid neoplasm. Therefore, the disease was considered idiopathic HES (IHES). The patient was treated with oral prednisolone 1 mg/kg daily. The eosinophil count dropped to 1.95 \( \times 10^9/L \) 1 week after the initiation of prednisolone therapy, and computed tomography scan showed absorption of the opacities in both lungs. That was also coupled with the disappearance of the skin lesions. Four weeks after the commencement of prednisolone therapy, the eosinophil count decreased to 0.04 \( \times 10^9/L \). The improvement in laboratory data was associated with disappearance of all symptoms, and the patient was discharged on a regimen of oral prednisolone. A follow-up plain X-ray film (Fig 3) taken 2 months later showed clear lung field and disappearance of all pulmonary opacities.

Comment

The criteria for the diagnosis of IHES, which was first defined by Chusid and associates [3] in 1975, have evolved throughout the years, because of the elucidation of the etiologies of some types of HES. Based on the consensus statement published by the International Workshop on Eosinophilic Diseases in 2006 [4], eosinophilic disorders are considered to fall into an umbrella category known as “hypereosinophilic syndromes” when an underlying allergic, parasitic, or other cause cannot be identified. The diagnosis of IHES under this classification scheme requires further exclusion of “myeloproliferative variants” and “lymphocytic variants” by testing for clonal abnormalities [5]. More recently, Simon and colleagues [6] proposed that the minimum duration of
eosinophilia criterion (6 months) need not necessarily be fulfilled for the diagnosis of HES, because any delay in the treatment of this progressive disease can put the patient at risk of irreversible organ damage. In our case, the eosinophil count increased progressively within 1 month and caused symptoms in lungs and skin of the thoracic wall. Because secondary etiologies were excluded and no clonal marker of myeloid or lymphoid neoplasm was identified, the diagnosis of IHES was made, and corresponding therapeutic interventions were initiated immediately.

All organ systems are susceptible to the effects of sustained eosinophilia, with cardiac (frequency of involvement, 58%), hematologic (58%), dermatologic (56%), neurologic (54%), and pulmonary (49%) manifestations being the most common [7]. Our patient had erythematous nodules on the right chest wall and several lung manifestations during the course of the disease. Although pleural effusion and lung opacities are among the common radiologic findings in HES [2], acute pneumothorax in one side followed by the same on the other side as the initial presentation has never been reported before.

What was the pathomechanism of pneumothorax in our patient? Secondary pneumothorax is caused by direct pleural damage or rupture of bullae in the setting of underlying lung disease, such as chronic obstructive pulmonary disease and postinflammatory fibrotic lesions (e.g., silicosis, chronic tuberculosis). In our patient, it is possible that the eosinophils, which infiltrated into the lung tissue and released cytotoxic substances, resulted in lung damage and subsequent bullae formation. It is also possible that pneumothorax is due to direct pleural injury by substances released by eosinophils, because the lesions were quiet near the pleura in both lungs (Fig 1B).

We have presented a case of IHES in a middle-aged man who presented with acute bilateral pneumothorax. The clinical symptoms and chest radiographs markedly

Fig 2. (A) Histopathologic examination of a biopsy taken from one skin lesion on the chest wall. Marked infiltration of eosinophils was seen in the fibroadipose tissue (hematoxylin-eosin, original magnification ×100). (B) Enlargement of boxed area in A; the fibroadipose tissue was edematous and contained eosinophilic infiltration, but no granuloma formation or signs of vasculitis (hematoxylin-eosin, original magnification ×400). (C) The bone marrow biopsy demonstrated increased proportion of eosinophils (arrowheads). The dark blue cells are lymphocytes or nucleated erythrocytes (hematoxylin-eosin, original magnification ×400).

Fig 3. Chest radiograph showed dramatic improvement after corticosteroid therapy.
improved after diagnosis and subsequent treatment with corticosteroids. Although IHES is a rare disease, it should be on the list of differential diagnoses of pneumothorax and migratory pulmonary opacities, and peripheral blood tests and bone marrow biopsy should be performed to confirm the diagnosis.

References

Aneurysm of the Pulmonary Vein: An Unusual Cause of Stroke
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This clinical report deals with a giant true pulmonary venous aneurysm, which was partially thrombosed. The overall incidence of pulmonary venous aneurysms is unknown, and they are reported only occasionally. We present the case of a previously healthy man with acute onset of ischemic cerebral stroke. The cause was a thrombus in a huge aneurysm of the left superior pulmonary vein. The patient subsequently underwent uncomplicated therapy for stroke, including thrombolysis followed by excision of the giant pulmonary venous aneurysm. As curative therapy we recommend complete resection of this rare entity.

A 59-year-old man presented with a first session of acute onset of right-sided hemiparesis and dysphasia. He had been previously healthy, had not experienced any trauma, and had no prior history of cardiovascular disease or vasculitis.

On admission, his vital signs and electrocardiographic findings were unremarkable, with sinus rhythm. The results of initial laboratory tests were within normal limits.

The patient was given thrombolytic therapy, and his neurologic symptoms resolved. The standard chest roentgenogram revealed a left parasternal mass. Evaluation included contrast computed tomography of the thorax, which showed a giant aneurysm with thrombus formation of the left superior pulmonary vein, including absence of perfusion to the left apical and apicoposterior segment 1 and 2 of the upper lobe (Fig 1). Trans thoracic echocardiography revealed normal left ventricular systolic and diastolic function and no abnormalities, especially no mitral valve stenosis or regurgitation.

On the basis of the imaging findings, we decided for resection of the thrombosed aneurysm after complete neurologic convalescence. Inasmuch as there was a persistent thrombus (Fig 1), we decided to perform the operation immediately; nevertheless, we had extracorporeal circulation circuit on standby because of the unknown tissue fragility. The patient underwent an uncomplicated left anterolateral thoracotomy. The findings at operation included a saccular pulmonary venous aneurysm. There...