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CASE REPORT

Esophagopericardial fistula, septic shock and intracranial hemorrhage with hydrocephalus after lung transplantation

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Abstract A 57-year old woman underwent lung transplantation for non-specific interstitial pneumonia. Primary graft dysfunction was diagnosed requiring continued use of extracorporeal membrane oxygenation (ECMO). Within three days she developed recurring hemothoraces requiring two surgical evacuations. After ECMO removal a series of complications occurred within four months: femoral thrombosis, persisting tachycardic atrial fibrillation, pneumopericardium with an esophagopericardial fistula and purulent pericarditis, septic shock, multiorgan failure and intracerebral hemorrhage with ventricular involvement requiring external ventricular drainage. Interdisciplinary management coordinated by the intensive care specialist, transplant surgeon and pulmonologist with various interventions by the respective specialists followed by intensive physical rehabilitation allowed for discharge home on day 235 post transplant. Subsequently quality of life was considered good by the patient and family.

Introduction

Lung transplantation (LTx) is a standard procedure for selected end-stage lung diseases. Complications such as

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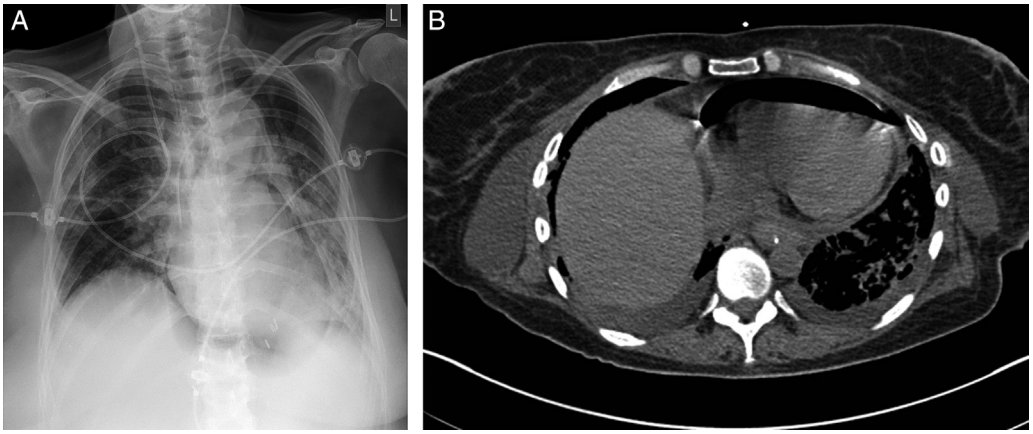

Figure 1 (A) Posterior–anterior view of chest radiograph with signs of pneumopericardium and left lower lobe opacification. (B) Chest CT showing pneumopericardium.

infection, rejection and malignancy contribute to the inferior survival rates of lung transplant recipients (LTR) as compared to other solid organs transplant patients. Due to polymedication, complications may present and respond to treatment differently and serial complications may be challenging for patients, relatives and healthcare professionals. We communicate here our most challenging case encountered in two decades of LTx.

Case report

A 57-year old woman received bilateral LTx for advanced non-specific interstitial pneumonia diagnosed by biopsy prior to transplantation. Her previous medical history was notable for a pulmonary sarcoidosis diagnosed 10 years ago and pandiverticulosis without infection. Mild pulmonary hypertension (PH) pre-transplant of 30 mmHg was known. Intraoperative clamping of pulmonary artery led to a mean pressure of 40 mmHg, so that ECMO was installed. In patients with PH requiring ECMO for transplant we routinely continue ECMO for weaning in the ICU setting. Postoperatively she remained ECMO-dependent due to a primary graft dysfunction. Within 24 h postoperatively she developed a left-sided hemothorax that needed surgical evacuation. The left atrium anastomosis was the source of the bleeding. Two days later a recurrence of the hemothorax occurred requiring surgical revision. These bleedings were facilitated by reduced coagulation due to heparin use required for ECMO. The improved respiratory situation allowed for removal of veno-arterial ECMO immediately after the second hematoma evacuation. A tracheostomy was performed due to expected extended ventilation requirements. An enteral feeding tube was installed endoscopically on postoperative day 4 in order to provide postpyloric feeding and medication. Gastroscopy showed some gastric erosions likely due to the nasogastric tube. On day 7 swelling of the right leg led to the diagnosis of thrombotic occlusion of the external iliac, common femoral and proximal femoral veins by duplex sonography requiring therapeutic anticoagulation. Thrombosis was considered a likely consequence of large-bore cannulation for ECMO.[1] A brief bronchoscopic evaluation for unexplained increased C-reactive protein of 76 mg/L on day 9 showed no evidence for anastomotic or infectious complications. For persisting atrial fibrillation amiodarone was started. Subfebrile temperatures were noted on day 11–15 with no obvious focus of infection from extensive sampling. Only recurrent gastroesophgeal reflux was detected, so antireflux measures were increased. Successful weaning led to decannulation on day 22 and transfer to the regular transplant ward two days later. On day 26 dyspnea, palpitations and anxiety revealed recurrent tachycardic atrial fibrillation requiring intermediate care admission for additional IV amiodarone loading doses. After a central venous line replacement the chest x-ray showed a pneumopericardium (Fig. 1A), which was confirmed by chest CT scan (Fig. 1B). Within 4 h the patient deteriorated rapidly due to septic shock leading to reintubation, circulatory support, hemofiltration and escalation of the antimicrobial treatment. Diagnostic evaluation revealed signs of reflux and candida esophagitis, esophagopericardial fistula with a visible 6 mm opening in the posterior left aspect of the esophagus requiring placement of a partially covered 12 cm self-expandable esophageal stent. A percutaneous pericardial tube drainage/flush system was inserted showing purulent pericardial effusion and retracheostomy was performed. Enterococcus faecium was cultured in the endobronchial wash from the apical left lower lobe infiltrate and the same organism was repeatedly cultured from the pericardial effusion during the first 5 days of the 3 week drainage period. Intravenous teicoplanin and caspofungin were given and daily pericardial taurolidine instillations were performed. Stent and tracheostomy removal were possible on day 43 and 48 post-transplantation respectively. Visible persistence of the fistula orifice in the esophagus required immediate closure with an Over-The-Scope-Clip (Fig. 2). Transfer from ICU to the regular transplant ward was possible on day 56. Intermittent dialysis was still required until day 63. Due to critical illness polyneuropathy/myopathy intensive inpatient rehabilitation was necessary.

On day 107 the patient showed no arousal and response to physical stimulation. The CT scan of the brain revealed a bleeding in the basal ganglia of the left side that had ruptured into the ventricular system with subsequent occlusive hydrocephalus (Fig. 3). This complication occurred on correctly dosed and monitored therapeutic IV heparin.

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