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CASE REPORT

The successful management of a Bronchoesophageal fistula after lung transplantation: a case report

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Summary

We describe an unprecedented, disastrous complication after bilateral lung transplantation (BLT), a bilateral bronchial dehiscence with a right bronchoesophageal fistula leading to life-threatening septic shock. We also report the successful endoscopic management of this complication by double stenting and stress the efficacy of the multidisciplinary approach to this critical case.

Introduction

Anastomotic airway complications are a major source of mortality and morbidity after lung transplantation. Among the most challenging ones, bronchial dehiscence, which tends to occur early, and fistulas stand out. The management of airway complications is a critical issue. We describe a case of bronchoesophageal fistula, an unprecedented complication after lung transplantation, and its successful management by double stenting (bronchus and esophagus).

Case report

A 24-year-old woman underwent BLT under emergency priority for terminal respiratory failure with recurrent pulmonary infections and chronic colonization by pseudomonas aeruginosa in cystic fibrosis after 5-day veno-venous extracorporeal membrane oxygenation (ECMO) support. Intra-operatively, cardiopulmonary bypass between the right atrium and the ascending aorta, according to the habit of our center, was established because of the onset of hemodynamic instability after the first pneumonectomy. The extracorporeal support was stopped after the implantation of the second graft. The bronchial anastomoses were performed with 4-0 PDS™ II running suture. On postoperative day (POD) 1, the patient was extubated upon a regular routine fibrobronchoscopy (FBS). Despite her well-compensated liver function at transplantation, the early postoperative course was complicated by liver failure with grade IV encephalopathy, which was effectively treated. About

2 weeks later, dyspnea and fever appeared and chest X-ray showed bilateral pneumonia. Despite broad-spectrum antimicrobial therapy, respiratory distress got worse, so mechanical ventilation was started. Septic shock occurred with hemodynamic instability requiring vasopressor and inotropic agents and renal failure. Persistent air leaks were measured during mechanical ventilation. On POD 30, a FBS pointed out a bilateral anastomotic dehiscence, M3aD0S1f according to the MDS classification [1]: on the right side, it extended for about one-fourth of the circumference with a fistula between the membranous bronchus and the esophagus; on the left side, it involved the membranous bronchus for about one-eighth of the circumference. Thus, two bronchial self-expanding uncovered metallic stents (Ultraflex™, Boston Scientific Corporation), 20-mm-long × 10-mm-diameter on the right and 30-mm-long × 10-mm-diameter on the left, were placed to overlay the bronchial defects (Fig. 1); soon afterward, an esophageal self-expanding fully covered metallic 80-mm-long × 18-mm-diameter stent (Niti-S™; Taewoong Medical, Seoul, Korea) and a nasogastric tube were positioned (Fig. 2). Veno-venous ECMO with lung-protective mechanical ventilation was set up. Subsequent targeted antimicrobial therapy, continuous veno-venous hemofiltration (CVVH) and volemic resuscitation lead to hemodynamic optimization with no longer need for inotropic drugs. As the infective status was gradually solved, the patient was weaned first from ECMO support, which was stopped 14 days after its start, and then from mechanical ventilation. About 4 weeks after the stents placement, a contrast swallow showed some water-soluble medium seeping between the stent and the esophageal wall into the fistulous tract, without any communication with the right bronchus (Fig. 3). The bronchial stents were removed 37 days after their positioning, upon endoscopic confirmation of the healing of the dehiscences. About 3 weeks later, the fistula was no longer detected by another contrast swallow (Fig. 4). Thus, the patient, who had been fed on enteral nutrition by the nasogastric tube up to that moment, gradually reintroduced oral feeding. The esophageal stent was removed 79 days after its placement. The patient was discharged from the hospital on POD 86. Ten months after the transplantation, follow-up FBS and contrast swallow proved the regularity of both the bronchial anastomoses and the esophageal transit, ruling out stenoses and fistulas. Two months later, the patient underwent liver transplantation for terminal hepatic failure in cystic fibrosis without any respiratory complications. Eighteen months after the lung transplantation, she is alive with good pulmonary and liver function.

Discussion

Anastomotic airway complications after lung transplantation have a reported incidence of 1.6–33% with an associated mortality rate of 2–4% [2] and a described prevalence of 5–20% [3]. The wide range of incidence and prevalence estimates can be attributed to the lack of a universally accepted grading system and surveillance program. Despite improvements in their management, the prevalence of airway complications has remained constant over the last decade, with a deleterious long-term impact [3]. Risk factors are controversial: ischemia of the donor bronchi due to their length and devascularization at harvest, size mismatch between donor's and recipient's airways, surgical technique, microbial contamination of the operative field, post-transplant infections, mechanical ventilation, immunosuppressant agents [1].

Fistulas between the bronchial tree and the surrounding structures are the result of bronchial dehiscence. In literature, three types of fistula after lung transplantation have been described: bronchopleural fistulas, bronchomediastinal fistulas, and bronchovascular fistulas [2]. Instead, no

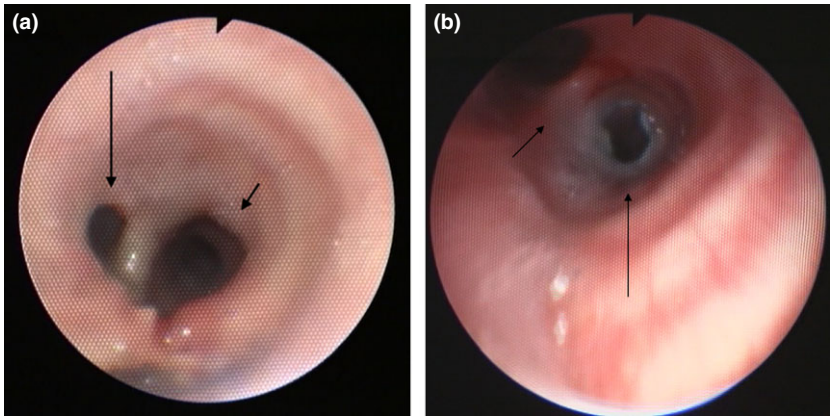

Figure 1 Bronchoscopic images: (a) of the right bronchial anastomotic dehiscence (long arrow) and the right graft main bronchus (short arrow); (b) of the right bronchial stent covering the dehiscence (the long arrow indicates the stent, the short one the main carina).

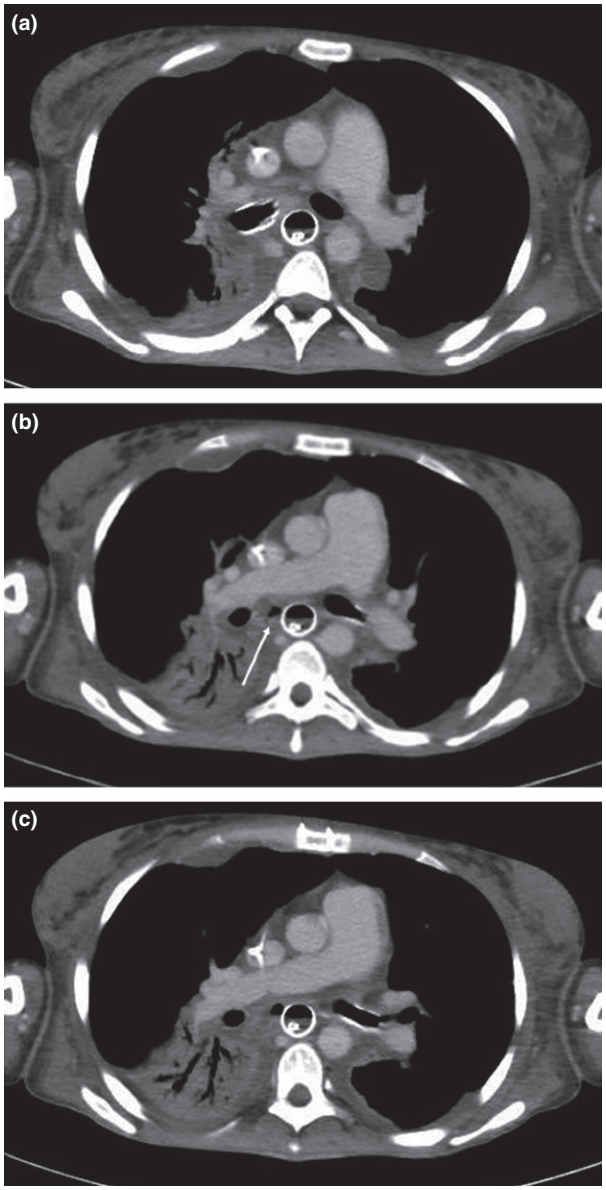

Figure 2 Contrast-enhanced CT showing: (a, c) the right and left bronchial stents, respectively, the esophageal stent and the nasogastric tube inside the esophageal stent; (b) air in the fistulous tract (arrow), excluded by the stents.

cases of post-transplant bronchoesophageal fistula have been mentioned.

The management of bronchial dehiscence after lung transplantation depends on the extent of the defect and the patient's conditions. The nature of interventions, if necessary, has shifted from surgical repair to endobronchial therapies, like the positioning of self-expanding metallic stents and silicone stents [3] or, rarely, the application of growth factors, autologous platelet-derived wound healing factors, and cyanoacrylate glue [2].

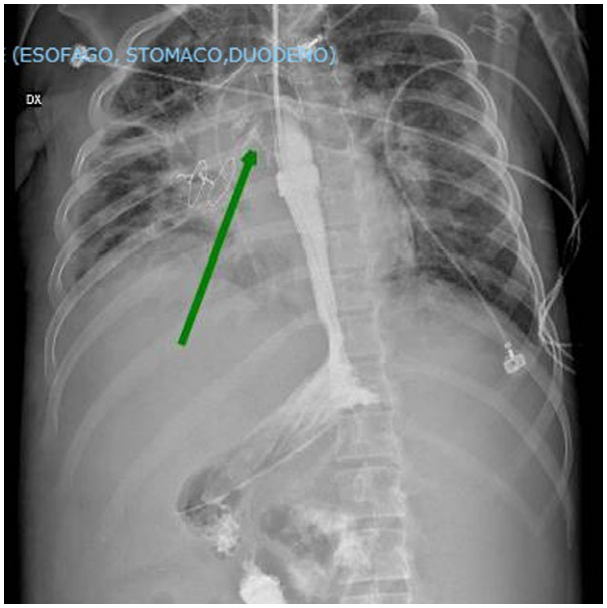

Figure 3 Contrast swallow performed with both the bronchial and esophageal stents still in place: some water-soluble medium appears inside the fistulous tract.

Malignant tracheo and bronchoesophageal fistulas as a complication of cancer arising in the esophagus, lung, or mediastinum are not so uncommon. Also in this situation, the endoscopic approach represents the favorite therapeutic strategy, including esophageal stenting, tracheobronchial stenting, and the simultaneous stenting of both the esophagus and the airway, while surgery is preferably avoided [4].

Benign not transplant-related bronchoesophageal fistulas in adults are rare and occur as a consequence of inflammatory disorders, foreign body or caustic ingestion, and congenital anomalies [5]. Again, endoscopic treatments such as the application of fibrin glue, sodium hydroxide, and acetic acid, the use of endoclips [6], laser therapy, and stenting [7] are preferred to surgery.

Chest tubes emerge from both the literature and common clinical practice as a useful tool for the conservative management of collections or air leaks due to esophageal or bronchial fistulas [8–10]. In the present case anyway, after the stents placement and the setup of veno-venous ECMO with lung-protective ventilation, neither significant collections nor air leaks were detected, so chest tubes would have not had any reasonable role.

Our patient was at high risk of postoperative complications because of her poor conditions at transplantation, which was performed under emergency priority after 5-day veno-venous ECMO support, and because of the chronic microbial colonization of her respiratory tract. We suppose that the esophageal perforation came first and the bronchial

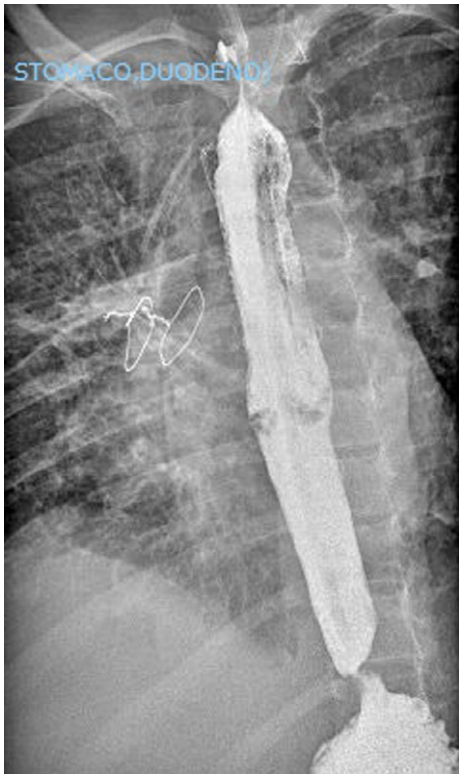

Figure 4 Contrast swallow performed with the esophageal stent still in place: there are no signs of the bronchoesophageal fistula.

dehiscence was the result of the exposure of the anastomosis to the esophageal leakage. The etiology of the esophageal perforation is not clear: we cannot rule out neither the devascularization of the esophageal wall during the preparation of the bronchial stump and tissue cauterization, nor the passage of a stitch through the esophageal wall while performing the bronchial anastomosis; the transesophageal echocardiography probe might have played a role too.

The prompt diagnosis of the bilateral bronchial dehiscence and of the right bronchoesophageal fistula and the multidisciplinary management of this complication and its critical consequences allowed the patient to survive an otherwise lethal situation. The technique of double stenting, borrowed from oncological palliation, proved to be effective at solving a previously unheard post-transplant complication, without any negative long-term impact.

Authorship

SC: wrote the manuscript. AL and VC: performed the transplant. PR and GBDD: placed the esophageal and bronchial stents, respectively. EB, AA, DP, MZ, MG and PP: were involved in the clinical management of the case. MC: was involved in the clinical management of the case and revised the manuscript.

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