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Case report

MANAGEMENT OF BRONCHOPLEURAL FISTULA WITH DIFFERENTIAL LUNG VENTILATION AND EXTRACORPOREAL MEMBRANE OXYGENATION

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Abstract

Bronchopleural fistulas (BPF) represent an unusual but clinically significant condition in pediatric patients. We report the case of a 15-year-old adolescent who developed septic shock and multiple organ failure secondary to a methicillin-resistant *Staphylococcus aureus* infection, complicated by necrotizing pneumonia and high-output BPF. The patient was supported with veno-venous extracorporeal membrane oxygenation and differential lung ventilation using a double-lumen endobronchial tube. This strategy enabled the control of positive pressure in the affected lung, promoting fistula closure and subsequent respiratory recovery. This case highlights the value of a multimodal approach in complex situations, emphasizing the synergistic use of extracorporeal membrane oxygenation (ECMO) and differential ventilation as a therapeutic option for refractory BPF in pediatric patients.

Key words: intensive care units, pediatrics, invasive ventilation, extracorporeal membrane oxygenation, one-lung ventilation

Resumen

Manejo de fístula broncopleural con ventilación pulmonar diferencial y oxigenación por membrana extracorpórea

Las fístulas broncopleurales (FBP) representan una condición inusual pero clínicamente significativa en pacientes pediátricos. Presentamos el caso de un adolescente de 15 años con *shock* séptico y falla de múltiples de órganos secundario a infección por *Staphylococcus aureus* meticilino resistente, que desarrolló neumonía necrotizante complicada con FBP de alto débito y recibió soporte con oxigenación con membrana extracorpórea (ECMO) veno-venoso y ventilación pulmonar diferencial mediante tubo endobronquial doble lumen. La estrategia permitió el control de la presión positiva del pulmón comprometido, favoreciendo el cierre de la fístula y la recuperación respiratoria del paciente. Este caso resalta el valor del abordaje multimodal en situaciones complejas, destacando el uso sinérgico de ECMO y ventilación diferencial como opción terapéutica en pediatría ante FBP refractarias.

Palabras clave: unidad de cuidados intensivos, pediatría, ventilación invasiva, oxigenación por membrana extracorpórea, ventilación pulmonar diferencial

Bronchopleural fistulas (BPF) represent an uncommon but concerning condition in pediatric patients. They are defined as a pathological communication between the main, lobar, or segmental bronchi and the pleural space. The most common etiology in this population is pulmonary resection, although they may also occur in the context of severe infections, neoplasms, and other less frequent pulmonary conditions[1]. Initial management of these entities is based on supportive measures, including pleural drainage with or without controlled suction, optimization of nutritional status, targeted treatment of the underlying cause, and adjustment of ventilatory

support, particularly in patients receiving mechanical ventilation[2]. Reducing airflow through the fistula is essential to promote spontaneous closure and limit associated complications. The presence of a BPF is associated with prolonged hospitalization, increased morbidity, and delayed recovery of pulmonary function.

Clinical case

We present the case of a 15-year-old male patient with no relevant past medical history, referred to the pediatric intensive care unit at Hospital Italiano de Buenos Aires due to septic shock with multiorgan failure secondary to septic arthritis and infectious endocarditis, with isolation of methicillin-resistant *Staphylococcus aureus* (MRSA). A trauma-related wound to the right knee was associated with the onset of this event.

Upon admission, the patient was under deep sedation and analgesia, on invasive mechanical ventilation, with poor perfusion and high-dose inotropic support requirements. A diagnosis of tension pneumothorax was made, and a right-sided chest tube was inserted, leading to hemodynamic improvement. In addition, he presented with cellulitis in the right knee and a surgical wound with purulent discharge.

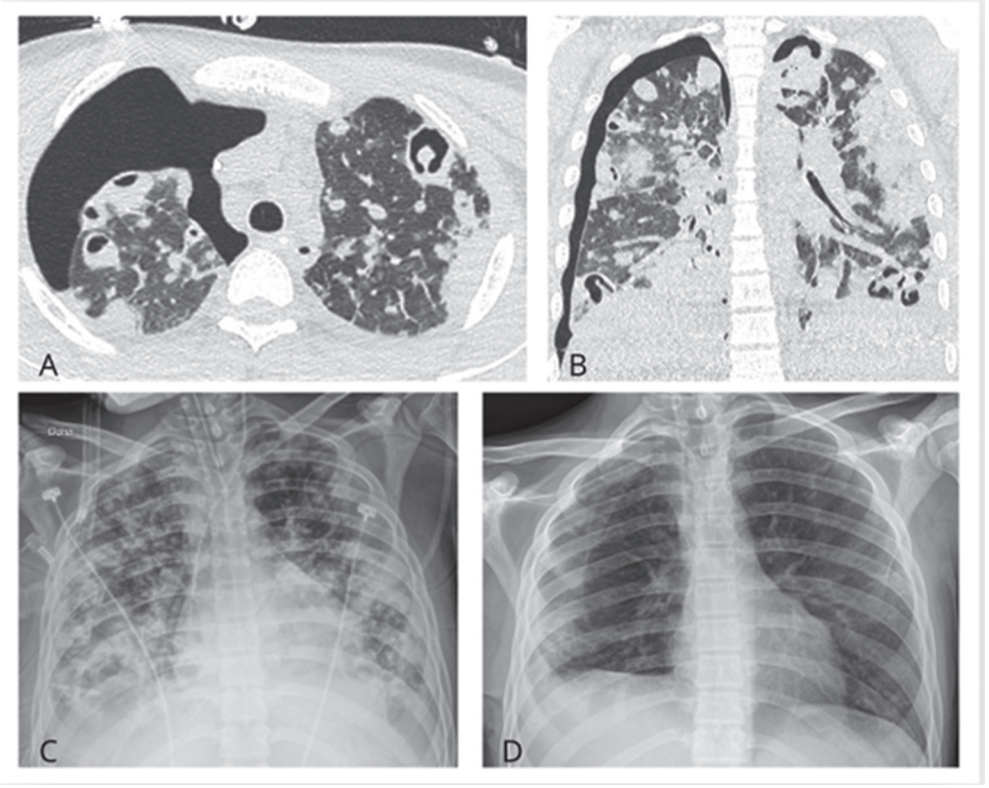
Key complementary studies included:

Doppler ultrasound of the right lower limb: femoral deep vein thrombosis.

Chest computed tomography (CT) scan: findings consistent with bilateral necrotizing pneumonia, residual right pneumothorax, and laminar pleural effusion. Presence of a right pleural drainage tube (Fig. 1). Cultures: blood, endotracheal aspirate, synovial fluid from the right knee, and pleural fluid were all positive for methicillin-resistant *S. aureus*.

During the clinical course, the patient developed severe respiratory distress associated with pulmonary hemorrhage and a high-output pleural fistula, secondary to necrotizing pneumonia. Due to refractory hypoxemia and respiratory acidosis despite conventional ventilatory

Figura 1 | A: Axial chest CT at admission showing bilateral cavitary lesions, ground-glass opacities, areas of consolidation and right-sided big pneumothorax. B: Coronal chest CT at admission demonstrating extensive bilateral parenchymal involvement with cavitation, consolidation and right-sided pneumothorax. C: Chest X-ray at admission showing diffuse bilateral opacities. D: Chest X-ray at discharge with marked improvement in lung aeration and resolution of most opacities



support, venovenous extracorporeal membrane oxygenation (ECMO) was initiated on the sixth day of hospitalization using a double-lumen cannula (Avalon Elite, Maquet, Rastatt, Germany®) inserted into the right internal jugular vein. It is noteworthy that an inferior vena cava filter was placed due to the femoral deep vein thrombosis and the initial contraindication to anticoagulation caused by the pulmonary hemorrhage.

Given the persistence of pneumothorax and air leak, evidenced by continuous bubbling in the water seal chamber, a second chest tube was inserted without resolution. Lung compliance was asymmetrical between the right and left lungs due to the parenchymal damage caused by infection. In this context, after 72 hours of ECMO support, differential lung ventilation (DLV) was initiated using a double-lumen endobronchial tube to individualize positive end-expiratory pressure (PEEP) and tidal volumes (VT) for each lung. This strategy allowed airway pressures to remain below the critical threshold for fistula reopening, promoting its closure.

As part of respiratory management, serial bronchoscopic lung toilettes were performed to evaluate hemorrhagic status, remove clots and blood remnants, apply topical tranexamic acid, and verify double-lumen tube positioning.

During the 11 days of differential lung ventilation, ventilator settings were progressively adjusted according to each lung's clinical course (Table 1). Initially, the left lung was ventilated in volume control (VC) mode with a VT of 50 milliliters (ml) and a PEEP of 10, while the right lung received 20 ml VT with a PEEP of 7. As pulmonary mechanics improved, tidal volumes were gradually increased (100 ml in the left lung and 70 ml in the right), and PEEP in the right lung was reduced to 5, always maintaining protective parameters. Once a total VT of 4 ml/kg was achieved, the patient was transitioned to a conventional orotracheal tube and switched to pressure control mode at 18/5 cmH_2O, achieving a VT of 190 ml while preserving a protective strategy without reopening the bronchopleural fistula.

The patient was successfully weaned from ECMO after 16 days of support. A tracheostomy was later performed to facilitate ventilator weaning, leading to successful decannulation and discharge from the hospital.

Parental consent to publish the medical case report of the child in a scientific journal was obtained.

Discussion

This case report presents the successful use of a rarely employed therapeutic strategy in pediatric patients with high-output bronchopleural fistula: the combination of ECMO and differential lung ventilation via selective intubation with a double-lumen endobronchial tube. Although unconventional in the pediatric population outside of the surgical setting, this approach constitutes a feasible alternative in selected cases.

In our patient, this strategy was implemented due to the inability to maintain spontane-

Tabla 1 | Arterial blood gas values and differential mechanical ventilation settings for the right lung and left lung on day 0, day 2, day 7, and day 10, showing respiratory evolution and extracorporeal membrane oxygenation parameters

	Day 0		Day 2		Day 7		Day 10	
Ventilator setting	RL	LL	RL	LL	RL	LL	RL	LL
Tidal volume (mL)	20	50	26	70	45	80	90	120
PEEP (cmH_2O)	7	10	7	10	5	10	5	10
Respiratory rate (bpm)	10		10		10		10	
Inspiratory time (sec)	0.9		0.9		0.9		0.9	
FiO_2 (%)	40		40		40		40	
Arterial blood gas								
pH	7.47		7.44		7.44		7.46	
pCO_2 (mmHg)	50		40		43		38	
HCO_3 (mmol/L)	36		29		28		27	
pO_2 (mmHg)	50		97		94		75	
Sat. O_2 (%)	96		92		95		94	
ECMO	2600 rpm - Flow 3.5 L/min - Sweep gas flow 6-8 L/min							

RL: right lung; LL: left lung; FiO_2: fraction of inspired oxygen; ECMO: extracorporeal membrane oxygenation; PEEP: positive end-expiratory pressure

ous ventilation in the setting of persistent air leak. The reduction of transpulmonary pressure in the affected lung, facilitated by selective intubation, promoted controlled atelectasis that favored fistula closure, while ECMO ensured adequate systemic ventilation and oxygenation. Among the main challenges encountered were fluctuations in ECMO flow, attributed to issues with the positioning of the double-lumen cannula. In addition, the presence of abundant clots and thick secretions prevented the withdrawal of mechanical ventilation, necessitating serial bronchial toilette procedures to clear both the endotracheal tube and the main airway.

Bronchopleural fistulas are a serious complication, typically secondary to pulmonary resection, infectious necrosis, pneumothorax, or tuberculosis[3]. Most resolve with conservative treatment through pleural drainage with or without suction systems[4], aimed at promoting spontaneous closure and preventing lung collapse, with reported spontaneous resolution rates of up to 80% in some series. Persistent air leaks and lack of adequate lung re-expansion predict the failure of conservative management and prompt consideration of more invasive interventions. In such cases, ventilatory strategies should focus on minimizing airflow through the bronchopleural fistula, achieved by reducing inspiratory pressure and tidal volume and employing low PEEP levels[1]. In refractory cases, ECMO emerges as a key therapeutic tool, ideally in the "awake ECMO" modality to avoid invasive mechanical ventilation. On the other hand, differential lung ventilation is a viable option for managing severe unilateral lung injuries[5], allowing functional isolation of the affected lung and promoting fistula closure.

Evidence on the combined use of ECMO and differential lung ventilation in the pediatric population is limited and consists primarily of isolated reports in surgical contexts. Our case is consistent with previously reported experiences[6,7]. As in those cases, the application of differential ventilation allowed for reduced pressure in the injured lung and, together with ECMO support, facilitated spontaneous fistula closure.

This case demonstrates that in complex and uncommon situations, such as high-output bronchopleural fistulas in pediatric patients, the combination of advanced strategies can be key to achieving a favorable outcome. The integration of ECMO and differential lung ventilation proved effective for pulmonary recovery. Appropriate patient selection, frequent endoscopic evaluations, and a multidisciplinary approach are essential for safe and effective implementation.

Our report aims to contribute to the literature by providing evidence on the application of combined strategies in the management of severe respiratory complications and highlights the importance of individualized treatment in critical care scenarios.

Conflict of interest: None to declare

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