CASE REPORT

Extracorporeal Membrane Oxygenation in Severe Pulmonary Forms of Leptospirosis: A Report of Two Cases

Suvadeep Sen[1], Alaukik Goyal[2], Vaishali Lokhande[3]

Abstract

Pulmonary involvement in leptospirosis is common. Severe pulmonary forms of leptospirosis (SPFL) carry high mortality. We report two cases of an otherwise healthy adult male from the western suburbs of India, admitted with severe pulmonary hemorrhage with extremely poor oxygenation. Veno-venous extracorporeal membrane oxygenation (VV-ECMO) was used as the last-rescue life-saving measure. Both the patients showed good pulmonary recovery within 2 weeks. Despite having thrombocytopenia, we experienced lesser bleeding complications requiring transfusions during the extracorporeal membrane oxygenation (ECMO) period.

Keywords: Acute respiratory distress syndrome, Extracorporeal membrane oxygenation, Leptospirosis, Tropical infections.

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Introduction

Leptospirosis is one of the emerging tropical infections that have significant morbidity and mortality.[1] Severe pulmonary forms of leptospirosis include severe pulmonary hemorrhage syndrome (SPHS) and acute respiratory distress syndrome (ARDS) and are associated with mortality up to 60%.[2] Treatment of SPFL is majorly supportive, apart from antimicrobials recommended for the leptospirosis. Evidence of use of ECMO in clinical literature is sparse. In this case series, we will discuss, two of our recent cases with SPFL, presented in extremely moribund conditions and treated successfully with ECMO. In the same opportunity, we shall discuss the potential role of this extracorporeal support system in this extreme form of this disease with a brief discussion of the existing literature.

Case 1

A 17-year-old boy was admitted with high-grade fever, vomiting, loose motion for 3 days, and breathlessness for 1 day. On admission, he was alert and oriented, but hemodynamically unstable, requiring noradrenaline support. He was breathless and hypoxic and hence was started on non-invasive ventilation (NIV). His urine output was low and was having high anion gap metabolic acidosis (HAGMA). 2D echocardiography showed normal ventricular function. On NIV, he developed right-sided pneumothorax, which worsened hypoxia; hence, intercostal drainage (ICD) was inserted, and he was intubated and started on lung-protective ventilation (LPV). During intubation, bleeding was noted from the trachea. He remained persistently oliguric, with increasing metabolic acidosis, hence, was started on continuous renal replacement therapy (CRRT). A decision was made to start VV-ECMO as rescue therapy. Gradually hemodynamic stabilization was achieved, and renal function improved. He was completely weaned off from ECMO on day 9 and was discharged from the hospital on day 16.

Case 2

A 46-year-old gentleman, farmer by profession, was admitted with a history of fever for 4 days and increasing breathlessness for 1 day. On arrival, he was severely hypoxic and hemodynamically unstable. He was intubated immediately and started on LPV. On attempting prone ventilation, his oxygenation worsened and hypercarbia increased, so, the decision to start VV-ECMO was taken as emergency-rescue maneuver. He also required CRRT during initial days of ECMO. Extracorporeal membrane oxygenation was weaned off in the next 13 days, and the ventilator was discontinued after 17 days. He showed slow recovery of kidney function, and he continued to require intermittent hemodialysis after discharge from the hospital for a month, following which renal function gradually returned to normal.

Clinical parameters, laboratory investigations, and ECMO details of both the patients have been mentioned in the supplementary material (Tables S1 to S3), respectively, and evolution of chest X-rays has been shown in Figures 1 and 2.

Discussion

Leptospira is a zoonotic disease and is more prevalent in tropical climate, rainy season, and urban slum areas with poor sanitation. Humans acquire infection when abraded skin or mucous membranes encounter water or soil contaminated with excreta of infected hosts.

Weil's syndrome is the severe form of leptospirosis, occurring in 5–10% of cases and may have multisystem organ involvement, including, jaundice, renal failure, and hemorrhagic complications.

[1,2]Department of Critical Care, Apollo Hospitals, Mumbai, Maharashtra, India

[3]Department of Internal Medicine, Apollo Hospitals, Mumbai, Maharashtra, India

Corresponding Author: Alaukik Goyal, Department of Critical Care, Apollo Hospitals, Mumbai, Maharashtra, India, Phone: +91 8530410945, e-mail: alaukikgoyal@gmail.com

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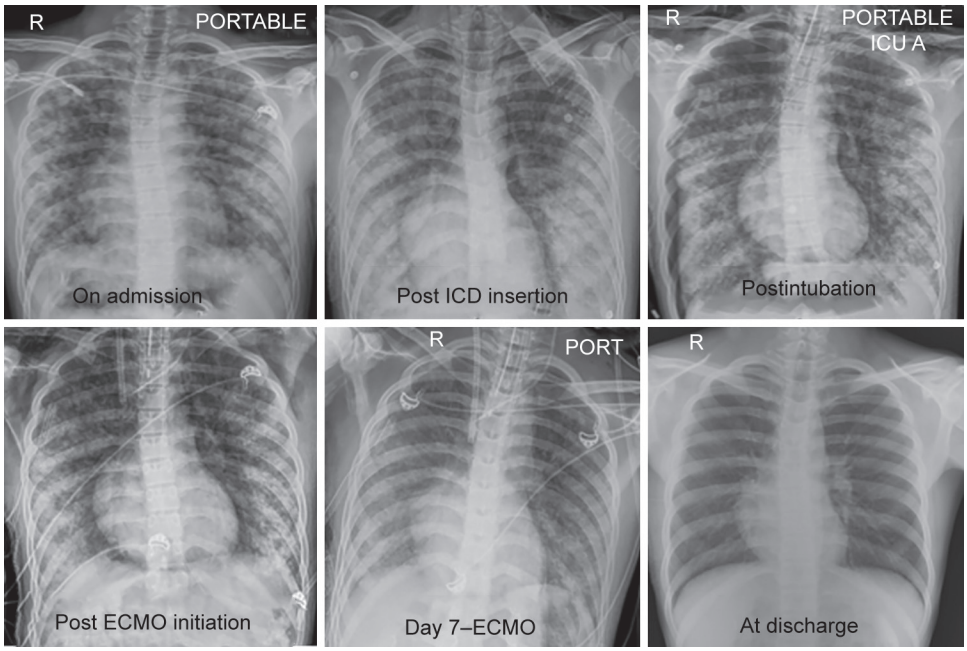

Fig. 1: Evolution of chest X-ray in patient 1

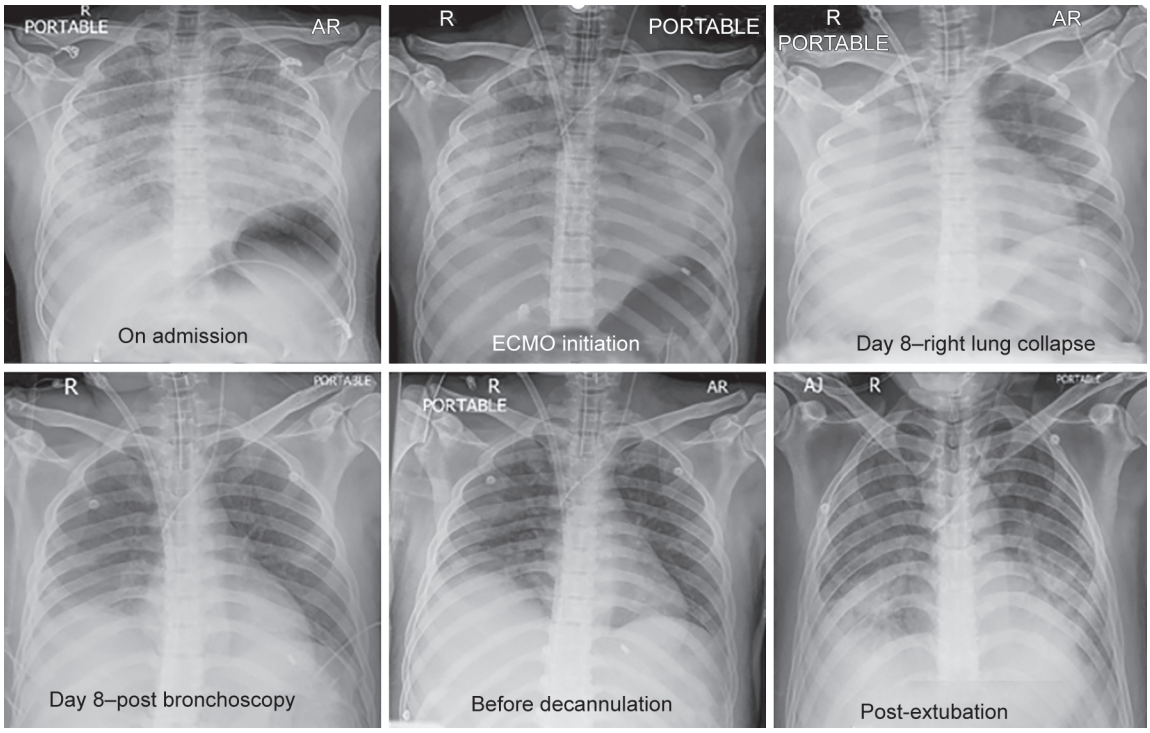

Fig. 2: Evolution of chest X-ray in patient 2

Pulmonary involvement of the disease is common (up to 70% of cases)[3] and sometimes a predominant clinical feature that can progress to pulmonary hemorrhage (SPHS) and ARDS. Treatment for SPHS is predominantly supportive along with the use of antimicrobials and invasive mechanical ventilation if required.

As per extracorporeal life-support organization guidelines, VV-ECMO is indicated in acute respiratory failure, where expected mortality with conventional measures is higher than 80%, and there is good reversibility of the primary disease.[4] This supports gas exchange and minimizes the damage induced by mechanical ventilation, and helps the lungs to recover. Use of ECMO in pulmonary hemorrhage is challenging, as systemic anticoagulation required for ECMO-circuit patency may increase risk of further alveolar bleeding.

In our discussion, both the patients had multiple-organ involvement and pulmonary hemorrhage with high SOFA scores at admission (Supplementary Tables S1 and S2), suggesting predicted mortality ≥95.2%. Pre-ECMO Murray's score was

3.8 and 3.5 in cases 1 and 2, respectively. Regarding ECMO management, standard insertion and maintenance techniques were used (Supplementary Table S3). Heparin was used for anticoagulation of ECMO circuit with target ACT 140–160. There was no ECMO circuit-related complication in either of the patients. Our transfusion target was to keep hemoglobin >70 gm/L and platelet >50 × 10^9/L in the presence of bleeding during ECMO maintenance and accordingly our transfusion requirement matched the transfusion practices in other centers[5] (total 5 RBC transfusions in a total 22 ECMO days in 2 patients = 0.11 units of RBC/day/patient). Both the patients showed good pulmonary recovery with time that was established both clinically and radiologically (Figs 1 and 2).

Use of ECMO in severe pulmonary form of leptospirosis was reported very few in clinical literature. Going through the medical literature published in PubMed-indexed journals, we could find 10 case reports so far (Table 1). In most of the cases, RRT was used along with ECMO,[2,6–9] which signifies frequent renal involvement in Weil's disease. Plasmapheresis and IV methylprednisolone was used in some of the cases as an immunomodulator with questionable benefit.[8,10] In all cases, ECMO was used as the last resort of rescue measures and was found to have high survival benefits.[11–14] Veno-venous form was used as the predominant ECMO modality. In our scenario, both the patients required RRT as renal support. Methylprednisolone was not used in any cases due to its controversial role in leptospirosis. Plasma exchange was not considered in our cases as its role is not proven, and hemodynamic instability on admission precluded its use. Both of our patients survived with complete recovery of organ function with time.

Conclusion

The present case report is important because our findings suggest the possibility of improving patient survival by using VV-ECMO in patients with leptospira pulmonary hemorrhage. To our knowledge, this is the first case reported from India. A good awareness is necessary to use ECMO as a potential modality in SPFL. A high index of suspicious, combined with timely appropriated supportive therapy, was key to patient survival.

Supplementary Material

All the Supplementary files are available at www.ijccm.org.

Table 1: Comparison of case reports using ECMO as therapy for SPFL

Mode	*P/F ratio*	*ECMO duration*	*Organ involvement*	*Adjuncts*	*Outcome*	*Country*	*Ref*
VV	57.7	183 hours	Cardiac arrest, jaundice	MARS	Recovery	UK	Arokianathan et al.[1]
VV	70.4	15 days	Kidney Liver Hypotension pericarditis	RRT Platelet 60 units 8 RBC, 4FFP	Discharged (Day 19)	Japan	Umei and Ichiba[2]
VV	<30	18 days	Hypotension ARF Liver	EACA infusion RRT Multiple platelet transfusions	Discharged (40 days)	USA	Pardinas et al.[6]
VV	77	8 days	Hypotension Liver Kidney	High-volume hemofiltration Several transfusions	Discharged (Day 28)	Chile	Cantwell et al.[7]
VV	51.8	17 hours	Hypotension Kidney Liver	CRRT Plasmapheresis 10 RBC, 12 gm fibrinogen Pulse methylprednisolone extracorporeal cytokine adsorbent filter	Died (Within 29 hours)	Germany	Ludwig et al.[8]
VV	62	6 days	Hypotension Kidney Liver	Pulse methylprednisolone Cyclophosphamide CRRT Hemoperfusion	19 days ICU	Philippines	Chavez et al.[9]
VV	74.2	8 days	Kidney Liver	IV MP 250 mg Plasmapheresis	Discharged (14 days)	Thailand	Chaikajornwat et al.[10]
VV			Retrospective analysis of 134 patients with leptospirosis 5 requiring VV-ECMO		4 survived	France	Delmas et al.[11]
VV	34	9 days	DIC Liver, ARF	8 RBC	Discharged (Day 20)	Laos	Héry et al.[13]
VV	50.5 42.4	9 days 13 days	Hypotension Kidney Liver Pancreas	5 PRBC 2 SDP for two patients	Discharged (16 days) (39 days)	India	Present report

ARF, acute renal failure; CRRT, continuous renal replacement therapy; DIC, disseminated intravascular coagulation; EACA, epsilon-aminocaproic acid; FFP, fresh frozen plasma; MARS, molecular adsorbent recirculating system; MP, methylprednisolone; PRBC, packed red blood cells; SDP, single donor platelet

Orcid

Suvadeep Sen https://orcid.org/0000-0001-8560-1208
Alaukik Goyal https://orcid.org/0000-0002-9061-4465
Vaishali Lokhande https://orcid.org/0000-0002-0909-2525

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