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Case report

Near-fatal negative pressure pulmonary oedema successfully treated with venovenous extracorporeal membrane oxygenation performed in the hybrid emergency room

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SUMMARY

We report a rare case of negative pressure pulmonary oedema (NPPE), a life-threatening complication of tracheal intubation. A 41-year-old obese man was admitted to a previous hospital for neck surgery. After extubation, he developed respiratory distress followed by haemoptysis and desaturation. The patient was reintubated and brought to our hospital where we introduced venovenous extracorporeal membrane oxygenation (ECMO) to prevent cardiac arrest, which is an unusual clinical course for NPPE. He returned to his routine without any sequelae. This is the first case report of NPPE successfully resolved with venovenous ECMO in the hybrid emergency room (hybrid ER), which is a resuscitation room equipped with interventional radiology features and a sliding CT scanner. Since the hybrid ER serves as a single move for patients where all necessary procedures are performed, it has the potential to lower the incidence of cannulation complications, beyond the delay in ECMO initiation.

BACKGROUND

Negative pressure pulmonary oedema (NPPE) develops as a result of upper airway obstruction and rapid increase in the negative intrapleural pressure due to attempts of intense inspiratory effort against an obstructed airway.[1 2] NPPE is a rare, but potentially life-threatening complication of general anaesthesia with tracheal intubation that occurs in 0.05%–0.1% of the cases.[1 3] Most patients with NPPE are treated with mechanical ventilation; thus, there are few reported cases requiring extracorporeal membrane oxygenation (ECMO).[4 5] Here, we present a near-fatal case of NPPE that was not diagnosed in a timely manner and was successfully treated with venovenous ECMO in the hybrid emergency room (hybrid ER), which is a novel resuscitation room equipped with a sliding gantry CT scanner and a fluoroscopic table with a self-propelled C-arm.

CASE PRESENTATION

A 41-year-old obese man (height, 176 cm; weight, 123 kg; body mass index, 39.7) with a medical history of hypertension, obstructive sleep apnoea (OSA) syndrome and cervical disc herniation (C5–C6) was admitted to a previous hospital for anterior cervical decompression and fusion surgery under general anaesthesia. He had no allergies but smoked one packet of cigarettes every day for 20 years. Preoperatively, his physical examination was unremarkable, and the vital signs were stable.

Brief timeline of clinical course during previous and our hospital is mentioned in table 1. General anaesthesia was induced by intravenous administration of propofol, fentanyl and rocuronium. Endotracheal intubation using an 8.5 mm internal diameter endotracheal tube was performed and he was maintained under general anaesthesia with sevoflurane, remifentanil and rocuronium. Surgery was uneventful, with a total of 2400 mL of the equilibrium solution was administered, the urine output was approximately 1000 mL and blood loss was 25 mL. The duration of the operation was 4.5 hours. Although extubation was performed after confirmation of complete consciousness and spontaneous breathing, he immediately developed respiratory distress, followed by haemoptysis and decreased peripheral oxygen saturation (SpO_2) to 23%. After reintubation, he was transferred to our hospital for angioembolisation of suspected bronchial artery bleeding.

TREATMENT

At the time of arrival to our hybrid ER, 3 hours had passed since reintubation. Ventilated with a bag valve mask, a pink-frothy fluid was observed coming out from the endotracheal tube and the patient was restless and agitated. Soon after admission, he was mechanically ventilated on volume control mode with tidal volume 6 mL/kg, positive end-expiratory pressure (PEEP) 12 cm H_2O, and FiO_2 100%, with the administration of intravenous midazolam and rocuronium. Despite increasing the PEEP to 15 cm H_2O, his SpO_2 was in the lower 80% range and venous blood gas demonstrated a pH of 7.17, $PaCO_2$ of 75 mm Hg, lactate levels of 51 mg/dL and a bicarbonate level of 26.6 mmol/L. He approached death due to bradycardia, hypoxia and hypercarbia. Since the echocardiogram showed a hyperdynamic left ventricle and he had no past medical history related to heart disease, we decided to introduce venovenous ECMO due to respiratory failure. Due to his recent neck surgery, a percutaneous right venous femoral cannula (21 Fr,

Table 1 Brief timeline of clinical course during previous and our hospital

Events	Time (hours)	Heart rate (/min)	RR (/min)	Blood pressure (mmHg)	SpO_2 (%)	Additional notes
Before intubation	12:30	72	16	160/100	99	Obese male in his 40s admitted to the previous hospital for neck operation.
After extubation	17:00	130	40	200/120	23	Developed respiratory distress after extubation. Reintubation was performed.
Departure from previous hospital	19:43	70	25	110/70	70	Transferred to our hospital for further treatment.
Hospital arrival	20:08	120	40	231/160	54	Pink, frothy fluid came out of the endotracheal tube and the patient was agitated.
Decision to introduce ECMO	20:20	42	25	127/90	82	Gradually became bradycardia. Approached death due to hypoxia and hypercarbia.
ECMO initiation	20:26	76	12	112/88	98	Improved rapidly with initiation of ECMO.
ICU admission	22:50	59	12	123/68	100	Diagnosis of NPPE was made. Became stable with ECMO.
Weaned from ECMO	13:12	63	16	122/70	100	Weaned from ECMO. Respiratory settings: PC–AC mode, FiO_2 45%, PEEP 20 cmH_2O, PIP 30 cmH_2O, RR 15/min. PaO_2/FiO_2 ratio >200.

ECMO, extracorporeal membrane oxygenation; NPPE, negative pressure pulmonary oedema; PC–AC, pressure-control and assist-control; PEEP, positive end-expiratory pressure; PIP, peak inspiratory pressure; RR, respiratory ratio.

CAPIOX (X), Terumo, Japan) for drainage and a left venous femoral cannula (16.5 Fr, CAPIOX (X), Terumo, Japan) for blood return were chosen. ECMO was set using a centrifugal pump (CAPIOX emergency bypass system, Terumo, Japan) and a membrane oxygenator (CAPIOX (LX), Terumo, Japan).

Since he was treated in the hybrid ER, the resuscitation room equipped with interventional radiology features and a sliding CT scanner (figure 1), the transfer of this seriously ill patient to an angiography suite for ECMO introduction was not required. Using fluoroscopy of the hybrid ER, he was placed on the ECMO without any complications within 18 min of his arrival to our hospital. His vital signs improved rapidly with the initiation of 4.4 L/min ECMO flow and mechanical ventilation was changed to lung-protective strategy. Bronchoscopy was performed which showed a pink, frothy fluid but no bloody sputum. Further, when the bronchoscope was inserted beside the endotracheal tube, there was severe swelling of the oral cavity and the pharyngolarynx was not visible. Later, using the advantage of the hybrid ER, a CT scan was performed without requiring bed transfer. The CT scan showed bilateral pulmonary congestion accompanied by air bronchogram and lesions with ground-glass opacity; these showed marked improvement compared with scans taken at the previous hospital (figure 2). There was no expanding haematoma near the trachea or the surgery wound. Therefore, he was diagnosed with NPPE due to upper airway swelling induced by the orthopaedic surgery. We set the therapeutic targets as follows: (1) SaO_2 ≥90% or PaO_2 ≥60 mm Hg

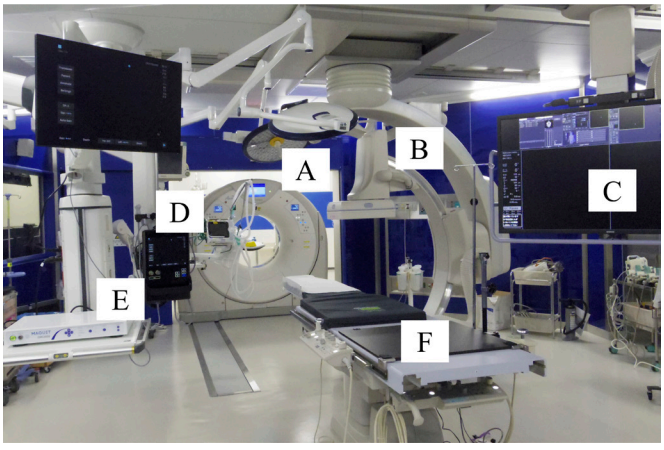

Figure 1 Photograph showing our hybrid ER. Equipped with a fluoroscopic table with a self-propelled C-arm and a sliding gantry CT scanner, the hybrid ER enabled us to perform multidisciplinary examinations and procedures such as fluoroscopy, ultrasonography, CT scan, angioembolisation and surgery on the same table without the need for patient transfer. (A) Sliding CT scanner, (B) self-propelled C-arm, (C) monitoring screen, (D) mechanical ventilator, (E) ultrasound equipment and (F) fluoroscopic table. hybrid ER, hybrid emergency room.

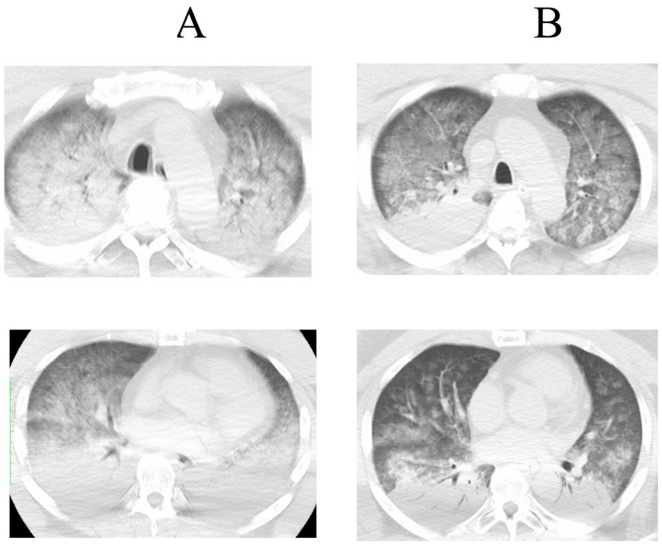

Figure 2 Comparison of the chest CT scan at the previous hospital (A) with the one taken at our hospital after venovenous ECMO initiation. (B) Both scans show bilateral pulmonary congestion accompanied by air bronchogram and lesions with ground-glass opacity which are consistent with NPPE. Although only 4 hours had passed between the two scans, a slight resolution of pulmonary congestion was confirmed after ECMO initiation at our hospital. ECMO, extracorporeal membrane oxygenation; NPPE, negative pressure pulmonary oedema.

and (2) pH ≥7.2. Since he was postoperative, we did not use heparin for anticoagulation. After confirming the improvement in the blood gas analysis and chest radiogram, he was weaned from the ECMO 17 hours after initiation. Since swelling of the oral mucosa and pharyngolarynx persisted, he was extubated successfully with the resolution of swelling on day 7. He was transferred to the previous hospital on day 9.

OUTCOME AND FOLLOW-UP

At 5 months postdischarge from our hospital, the patient conveyed through a telephonic interview that he had resumed work without sequelae.

DISCUSSION

We present a case of a 41-year-old obese man who underwent anterior cervical decompression and fusion surgery complicated by NPPE, a rare but fatal adverse event that involves the development of pulmonary oedema secondary to acute upper airway obstruction. Our case was unique since most of the reported NPPE cases resolved with oxygenation or reintubation[1 4]; however, we had to introduce ECMO to prevent the patient going into a cardiac arrest from severe hypoxia and hypercarbia. Further, using fluoroscopy of the hybrid ER, the patient was rapidly and safely placed on ECMO without transfer to an angiography suite.

NPPE, also known as postobstructive pulmonary oedema, is characterised by the formation of negative intrathoracic pressure in cases with upper airway obstruction, leading to severe hypoxemia and pulmonary oedema.[6] As a serious but rare complication, NPPE is estimated to occur in 0.05%–0.1% of postanaesthesia patients with tracheal intubation.[1 3] It is important to perceive the potential risk factors of NPPE. A review of NPPE cases reported 29 cases related to general anaesthesia of which, 10 cases involved head and neck surgery that may be related to tissue swelling of the upper airway.[1] On similar lines, our patient underwent neck surgery, anterior cervical decompression and fusion surgery, and showed marked swelling in the oral cavity. Further, he had several known risk factors of NPPE such as obesity, young age, current smoker and OSA[7–9]; thus, the possibility of developing NPPE was considered.

To prevent NPPE in high-risk patients after cervical spine surgery, there is an airway management protocol that is based on five clinical risk factors, which can reduce postoperative airway complications.[10] If there are any one or more risk factors, such as medical comorbidities, operative site, operative range, operative time and blood loss, patients should be kept intubated for at least overnight. Extubation is performed proceeding from the positive results of the cuff-leak test or permissive swelling of prevertebral soft tissue in lateral radiographs. These tests are repeated after 12 hours until extubation criteria are fulfilled. Using this protocol, since this patient had medical comorbidities, he would be extubated after confirming the results of testing the next day or later. NPPE will be prevented if such procedures are generally recognised.

The diagnosis of NPPE is usually not challenging if there are series of episodes such as a rapid onset of respiratory failure after extubation, the existence of upper airway obstruction, the presence of pink tracheal secretions, the radiological findings of pulmonary oedema and a normal cardiac function on echocardiogram. Given that our patient was a relatively healthy young man with a well-controlled perioperative fluid balance, no history of newly developed left heart failure on an echocardiogram, cardiogenic oedema was not considered. Additionally, the time course of the clinical and radiologic recovery was in agreement with the aetiology of NPPE. In our patient, misdiagnosis of NPPE as bronchial artery bleeding at the previous hospital delayed the initiation of correct treatment which exacerbated his symptoms. Further, the presence of haemoptysis may also have made the diagnosis difficult in this case; although, a retrospective study showed that diffuse alveolar haemorrhage was associated with 10% of NPPE cases.[11] Almost all NPPE cases reported previously resolved with supportive measures; hence, few cases have been reported which required ECMO for NPPE.[1 4 5] Therefore, it is important to make the diagnosis as quickly as possible and treat it with compatible methods.

This case is notable for utilising the advantages of the hybrid ER optimally; we could make the diagnosis in a timely manner and initiate treatment safely and seamlessly. Equipped with a fluoroscopic table with a self-propelled C-arm and a sliding gantry CT scanner, the hybrid ER enabled us to perform multidisciplinary examinations and procedures such as fluoroscopy, ultrasonography, CT scan, angioembolisation and surgery on the same table (figure 1). Based on the advantages shown in previous studies, unstable patients may benefit from not having to move to various locales in the hospital; severe complications sometimes occur during transportation, such as tube or ventilator dislodgement, monitoring failure or inadequate equipment and manpower when something unexpected occurs.[12–14] Therefore, the hybrid ER may prove advantageous since patients would be stabilised in a single location where all necessary procedures can take place. Further, a retrospective study demonstrated that a hybrid ER significantly reduced the incidence of complications of cannulation of ECMO.[15] Thus, we could initiate ECMO within 18 min of arrival in the hybrid ER without any complication which prevented him from going into a cardiopulmonary

Patient's perspective

I remember that I was very surprised to hear that I was about to die. I do not really remember the days I spent in Saiseikai Yokohamashi Tobu Hospital. My family told me that an uncommon but one of the best medical treatments was performed on me. I appreciate the efforts taken by the physicians and the hospital team. I have already resumed work without any sequelae. Now, I have quit tobacco and I am also trying to lose weight to enjoy my saved life.

Learning points

- Negative pressure pulmonary oedema (NPPE) is a rare, but potentially life-threatening complication of general anaesthesia with tracheal intubation.
- Although most patients with NPPE are well treated with mechanical ventilation, we were presented with a near-fatal case of NPPE that was not diagnosed in a timely manner but successfully treated with venovenous extracorporeal membrane oxygenation (ECMO).
- The hybrid emergency room (hybrid ER) is a novel resuscitation room which enabled us to perform multidisciplinary examinations and procedures on the same table without patient transfer.
- The hybrid ER has the potential to lower the incidence of cannulation complications, beyond the delay in ECMO initiation.

arrest. Meanwhile, there are several issues such as the implementation cost, complexity and indication.[16] The appropriate way to handle the new system in the clinical practice remains unknown. The further research is needed.

In conclusion, NPPE should be considered in case of acute pulmonary oedema after extubation which is a rare, but fatal complication when inappropriately managed. In our case, since a delay in the diagnosis and treatment exacerbated the NPPE, ECMO had to be introduced to prevent the patient from going into a cardiac arrest. Although it is invasive, ECMO is known to be one of the most effective support measures for respiratory failure[17] and makes recovery possible without any sequelae. Further, there is a possibility that through the use of the hybrid ER, a safer and faster ECMO can be initiated.

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