Case Reports

Negative Pressure Ventilation via Chest Cuirass to Decrease Ventilator-Associated Complications in Infants with Acute Respiratory Failure: A Case Series

Hilary Klonin BM BS, Brian Bowman MD PhD, Michelle Peters RRT, Parakkal Raffeeq MD, Andrew Durward MD, Desmond J Bohn MD, Jon N Meliones MD, and Ira M Cheifetz MD

Pulmonary and nonpulmonary complications of invasive positive pressure ventilation are well documented in the medical literature. Many of these complications may be minimized by the use of noninvasive ventilation. During various periods of medical history, negative pressure ventilation, a form of noninvasive ventilation, has been used successfully. We report the use of negative pressure ventilation with a chest cuirass to avoid or decrease the complications of invasive positive pressure ventilation in three critically ill infants at two institutions. In each of these cases, chest cuirass ventilation improved the patient's clinical condition and decreased the requirement for more invasive therapy. These cases illustrate the need for further clinical evaluation of the use of negative pressure ventilation utilizing a chest cuirass. [Respir Care 2000;45(5):486–490] *Key words: noninvasive ventilation, negative pressure ventilation, chest cuirass ventilation, secretion clearance, mechanical ventilation, extracorporeal membrane oxygenation, respiratory failure.*

Introduction

Traditionally, mechanical ventilator support for respiratory distress and respiratory failure has been invasive positive pressure ventilation (PPV). However, there have been periods of time, such as during the polio epidemic, when negative pressure ventilation (NPV) has been successfully used on a relatively large scale. In the past, NPV has not remained popular because of technical limitations of NPV and difficulty performing general patient care in the "iron lungs." In order to reduce iatrogenic complications of invasive mechanical ventilation, a renewed emphasis on noninvasive ventilation, including NPV, is surfacing.

Complications of intubation and invasive PPV are well documented.[1] Potentially serious airway complications that involve the endotracheal tube (ETT) include incorrect ETT placement, traumatic injury during intubation, ETT obstruction, and ETT displacement.[1–2] Prolonged intubation may cause sinusitis, nasal septum injury, subglottic stenosis, and the development of airway granulomas from repeated endotracheal tube suctioning.[1–3] Additionally, accidental extubations may result in considerable morbidity or even mortality.[4] Conventional positive pressure mechanical ventilation may lead to clinically important pulmonary complications, including secondary lung injury.[5–7] Invasive PPV has been associated with the development of hyaline membrane formation and the potential for subsequent increases in ventilatory support.[8–9] Patients receiving mechanical ventilation are also at risk for nosocomial pneumonia.[10–11]

SEE THE RELATED EDITORIAL ON PAGE 479

The potential complications associated with PPV extend beyond the respiratory system. Adverse sequelae to the

Hilary Klonin BM BS, Brian Bowman MD PhD, Michelle Peters RRT, Jon N Meliones MD, and Ira M Cheifetz MD are affiliated with the Department of Pediatric Critical Care Medicine, Duke Children's Hospital, Durham, North Carolina. Parakkal Raffeeq MD, Andrew Durward MD, and Desmond J Bohn MD are affiliated with the Hospital for Sick Children, Toronto, Ontario, Canada.

Ms Peters presented a version of this paper at the American Association for Respiratory Care OPEN FORUM during the 44th International Respiratory Congress, November 7–10, 1998 in Atlanta, Georgia.

Correspondence: Ira M Cheifetz MD, Duke Children's Hospital, Duke University Medical Center, Box 3046, Durham NC 27710. E-mail: cheif002@mc.duke.edu.

cardiovascular and neurologic systems may occur. Substantial levels of conventional mechanical ventilatory support may lead to cardiovascular compromise with decreased cardiac output and compromised oxygen delivery.[12] The sedation and analgesia required for patients to tolerate invasive ventilation can result in the need for prolonged ventilation because of oversedation.[13] Withdrawal symptoms may develop in certain patients and may require a lengthy tapering of opiates and/or benzodiazepines.

The many difficulties associated with intubation and invasive PPV have lead clinicians to explore other means of respiratory support. Noninvasive ventilation avoids the need for an artificial airway while allowing for improved communication, coughing, and swallowing. Oral feeding may also be better tolerated. Noninvasive ventilation can be administered by both positive and negative pressure and may require substantially less sedation than invasive ventilatory techniques.

One of the currently available negative pressure ventilators is the Hayek Oscillator (Breasy Medical Equipment Ltd, London, UK).[14–15] The Hayek Oscillator consists of a chest cuirass attached to a piston pump that provides NPV at both conventional and high-frequency rates. The baseline negative pressure is produced by a vacuum pump. The chest cuirass is a lightweight, flexible chest enclosure with foam rubber around the edges to provide an airtight seal over the chest and abdomen.

The Hayek Oscillator offers several modes, including noninvasive ventilation around a negative baseline, continuous negative pressure (the negative pressure equivalent of continuous positive airway pressure), and secretion clearance. The secretion clearance mode consists of oscillations around a negative baseline followed by an artificial "cough." This artificial cough has a prolonged inspiratory phase followed by a forced short expiratory phase. The secretion clearance mode in combination with high-frequency external chest wall oscillation has been shown to increase mucociliary clearance in a canine model.[16–17]

Negative pressure ventilation using a chest cuirass has been previously reported in the literature for various purposes, including ventilatory support for patients (1) after congenital heart surgery, (2) with neuromuscular disease, (3) during failed fiberoptic intubation, (4) after lung resection for bullous emphysema, and (5) during microlaryngeal surgery.[18–23] In this case series, we report additional uses of chest cuirass NPV in the neonatal population. These cases illustrate the use of noninvasive NPV to avoid or decrease the complications of PPV in three critically ill infants. In this series, NPV was utilized in separate instances to avoid intubation, avoid reintubation, and facilitate weaning from extracorporeal membrane oxygenation (ECMO).

Case 1

A 6-month-old male infant was transferred to Duke Children's Hospital from a local referring hospital with a three-week history of diarrhea and increasing respiratory distress. The infant developed worsening respiratory failure with peripheral oxygenation saturations of 80% on a fraction of inspired oxygen (F_{IO_2}) of 0.60. A chest radiograph revealed a lingular infiltrate. Bronchoscopy confirmed the diagnosis of *Pneumocystis carinii* pneumonia. The patient's provisional diagnosis by immunologic testing was X-linked γ globinopathy, and the patient was started on co-trimoxazole.

Upon arrival to the pediatric intensive care unit, the infant was in respiratory failure, with a respiratory rate of 60 breaths per minute, severe intercostal retractions, grunting, and nasal flaring. An arterial blood gas analysis indicated: pH 7.40, arterial partial pressure of carbon dioxide 38 mm Hg, arterial partial pressure of oxygen (P_{aO_2}) 137 mm Hg, arterial oxygen saturation 96% on face mask with F_{IO_2} of 1.0. Additionally, the patient was noted to have decreased perfusion with weakly palpable peripheral pulses and prolonged capillary refill.

In an attempt to decrease the work of breathing and improve oxygenation, the patient was initiated on continuous negative airway pressure (–6 to –8 cm H_2O) using a Hayek Oscillator. Shortly after initiating continuous negative pressure via chest cuirass, the patient's respiratory rate decreased to 40 breaths per minute. The F_{IO_2} was weaned from 1.0 to 0.80 while maintaining oxygen saturation above 95%. However, over the next several hours the patient's oxygen saturation again decreased to $< 90\%$ with the F_{IO_2} remaining at 0.80. The infant's ventilatory support was, therefore, changed to intermittent NPV with a ventilatory rate of 40 breaths per minute and peak inspiratory and expiratory pressures of –18 cm H_2O and –2 cm H_2O, respectively. On these settings, the patient had decreased work of breathing, as evidenced by decreased retractions, grunting, and nasal flaring. The F_{IO_2} delivered via face mask was weaned to 0.60 while maintaining oxygen saturation $> 95\%$. The infant did not develop a metabolic acidosis on arterial blood gas analysis, and lactic acid levels never exceeded 2.1 mmol/L.

The negative inspiratory pressure was maintained between –18 cm H_2O and –22 cm H_2O for three days, with continued respiratory improvement. Subsequently, the patient improved with routine supportive care. The infant had progressive normalization of respiratory effort, respiratory rate, and oxygenation. By day 10 of hospitalization, he was receiving 1–2 L/min of oxygen via nasal cannula. By hospital day 12, the patient no longer required supplemental oxygen. At that time, the infant received a bone marrow transplant for treatment of his immunodeficiency syndrome and was transferred from the intensive care unit.

He did not require further admission to the intensive care unit during his hospital course.

Case 2

A 4-month-old former 26-week premature male infant (birthweight 905 g) was admitted to a referring hospital after a respiratory arrest at his home. At the time of his arrest, emergency medical service personnel performed the initial intubation and resuscitation. The infant's medical history was notable for chronic lung disease (CLD) of prematurity and recurrent apnea.

At a local hospital, the infant experienced progressive respiratory deterioration requiring increasing ventilatory support and a trial of high-frequency positive pressure oscillatory ventilation. The initial chest radiograph revealed diffuse air space disease consistent with an intercurrent respiratory viral infection. For transfer the infant was placed on pressure control ventilation: ventilatory rate 40 breaths per minute, peak inspiratory pressure 33 cm H_2O, positive end-expiratory pressure 10 cm H_2O, mean airway pressure (P_{aw}) 21 cm H_2O, inspiratory time 0.7 seconds, and F_{IO_2} 1.0. The calculated oxygenation index [$(P_{aw} \times F_{IO_2} \times 100)/P_{aO_2}$] was 19. On transfer to Duke Children's Hospital, the infant was placed on pressure control/pressure support ventilation: ventilatory rate 25 breaths per minute, peak inspiratory pressure 37 cm H_2O, positive end-expiratory pressure 8 cm H_2O, pressure support 10 cm H_2O, inspiratory time 0.6 seconds, and F_{IO_2} 1.0. He was extubated after one week of conventional mechanical ventilation.

After extubation the infant was noted to have a bulbar palsy, with inability to swallow his secretions, as well as weakened cough and gag reflexes. The postextubation chest radiograph showed collapse of his left lung and right upper lobe (Fig. 1A). At this same time, clinical evaluation revealed an increased respiratory rate to 60 breaths per minute, subcostal retractions, and grunting. Because of the patient's history of CLD, the medical care team desired to avoid reintubation and PPV. Thus, continuous negative pressure of –30 cm H_2O was initiated in an attempt to decrease his work of breathing and to improve the atelectasis. Additionally, every 2–3 hours the infant was placed in the secretion clearance mode, with three minutes of oscillation at 600 cycles per minute and three minutes of "cough" at 60 cycles per second. In an effort to support his hypotonic upper airway, nasal continuous positive airway pressure of 6–10 cm H_2O was applied. During NPV, enteral feeding was successfully accomplished via a nasoduodenal tube. After two days the left lung re-expanded, and after an additional day the right lung expanded (see Fig. 1B). He was gradually weaned off NPV over the next two days. His lungs remained expanded, and he was transferred back to his previous care facility two days later.

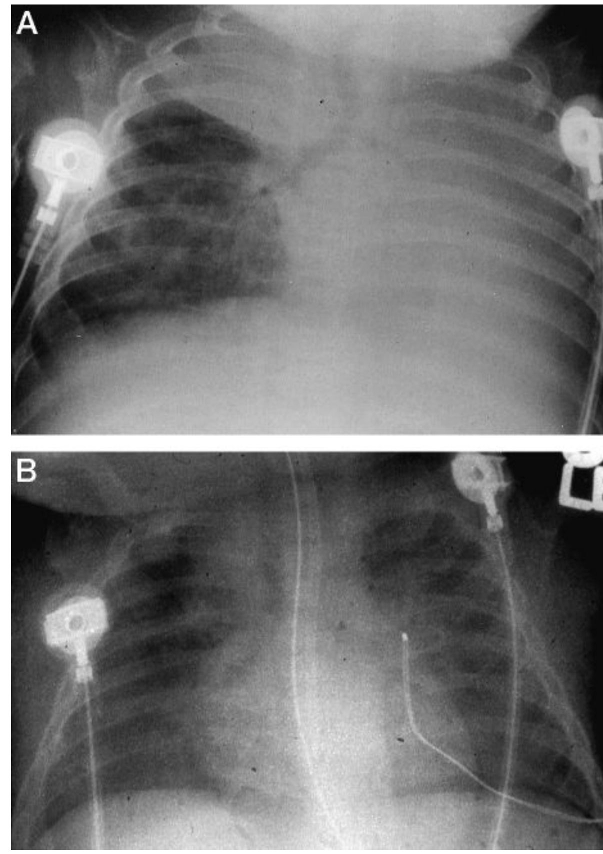

Fig. 1. A 4-month-old former 26-week premature male infant with history of chronic lung disease. The infant was intubated for an intercurrent viral illness. Figure 1A (upper) represents severe atelectasis after extubation. Figure 1B (lower) represents substantial improvement in the atelectasis after treatment with negative pressure ventilation, including use of the secretion clearance mode.

Case 3

A 16-month-old infant was referred to Toronto Sick Children's Hospital, having developed respiratory failure and a clinical diagnosis of bronchiolitis obliterans. On conventional mechanical ventilation, the infant developed progressive hypoxia and hypercarbia. Respiratory deterioration persisted despite a trial of high-frequency positive pressure oscillatory ventilation. Diffuse airway plugging and atelectasis was noted. Despite daily bronchoscopic alveolar lavage and administration of acetylcysteine (Mucomyst), DNAase, and exogenous surfactant, the atelectasis persisted. The patient's hospital course was complicated by the development of a life-threatening cardiac arrhythmia, further respiratory compromise and hemodynamic instability necessitating venoarterial ECMO. The oxygenation index immediately prior to ECMO was 60. During his ECMO course, he suffered a pulmonary hemorrhage on day 8. The patient was unable to be weaned off ECMO because of inadequate oxygenation despite high positive pressures on conventional mechanical ventilation. The pulmonary dynamic compliance remained at 0.35 mL/cm H_2O/kg.

After 18 days of ECMO the infant was placed on chest cuirass NPV at a rate of 30 breaths per minute with an inspiratory to expiratory ratio of 1:1, inspiratory pressure of –25 cm H_2O, expiratory pressure of 5 cm H_2O, and two cycles of secretion clearance every two hours. Additionally, the positive pressure conventional ventilator was set to deliver a pressure support of 10 cm H_2O and a positive end-expiratory pressure of 5 cm H_2O. These ventilatory settings produced good chest movement and did not interfere with the patient's hemodynamic status. While on the Hayek Oscillator, a large increase in secretions occurred, and the secretions were easily lavaged with routine endotracheal tube suctioning. After 24 hours the dynamic compliance doubled to 0.72 mL/cm H_2O/kg. After 48 hours the oxygenation index fell to 5, and the infant was successfully decannulated from ECMO. The patient remained on conventional mechanical ventilation for an additional three days and was subsequently weaned to nasal cannula oxygen. The infant was successfully transferred back to the referring hospital.

Discussion

Chest cuirass NPV may be used to avoid the potentially deleterious effects of invasive PPV. The potential beneficial effects of NPV can be divided into several categories, including reduced airway complications, improved pulmonary parenchymal inflation at reduced airway pressures, reduced cardiovascular compromise, decreased sedation requirements, and improved enteral nutrition. The cases described in this paper illustrate some of these potential benefits of chest cuirass NPV.

By eliminating the need for intubation, the potential airway complications associated with invasive PPV[1–4] can be completely avoided. Barotrauma, also associated with PPV,[5–7] can be avoided by the use of NPV. Not only does NPV avoid the detrimental effects that PPV can have on hemodynamics, NPV may actually improve the patient's hemodynamic status.[18,19,24,25] NPV may minimize sedation requirements, as it is generally well tolerated by most patients. Additionally, enteral feeding is usually well tolerated in patients treated with NPV alone.

Our first case in this series demonstrates the use of NPV to avoid intubation in a critically ill infant with *Pneumocystis carinii* pneumonia. Original assessments of patients with *Pneumocystis carinii* pneumonia requiring mechanical ventilation report a mortality of $>$ 80% in adults and 50% in children.[26–27] However, more recent reports indicate that the initiation of adjuvant therapy with corticosteroids, as well as earlier recognition of this disease, has led to an increase in the survival rate of patients with acquired immunodeficiency syndrome and respiratory failure secondary to pneumonia.[28–30] In the patient presented, steroids were felt to be contraindicated because of his congenital immune deficiency syndrome. In view of historically poor results with ventilation of severely immunocompromised patients, we wished to avoid intubation and PPV. In this patient, NPV indeed saved this child from the risks of ventilator-associated pneumonia, barotrauma, and possible need for inotropic support.

The second infant that we described had CLD of prematurity, or bronchopulmonary dysplasia, which has been well described as a complication of PPV.[31] Although reventilation for lung expansion was an option, this patient's lungs were already severely damaged and would have been vulnerable to further ventilatory trauma by PPV. The use of continuous negative pressure for alveolar recruitment combined with the physiotherapy mode for secretion clearance was effective in improving the patient's clinical course while avoiding PPV. This method has not previously been documented in the literature.

The patient in Case 3 was failing to show improvement despite maximal standard therapy. An oxygen index of $>$ 40 prior to ECMO has been associated with a mortality of $>$ 80%.[32] The patient's ECMO course was complicated by a pulmonary hemorrhage, emphasizing the need to remove this infant from ECMO as soon as possible. However, a pulmonary compliance of $<$ 0.6 mL/cm H_2O/kg has been associated with failure to wean from ECMO.[33] Since this patient's pulmonary compliance failed to improve despite maximal "conventional" therapy, it was felt that further options were limited. The secretion clearance mode on the Hayek Oscillator offered this child chest physiotherapy and the ability to mobilize secretions. The improved secretion clearance helped to increase the infant's pulmonary compliance from 0.35 mL/cm H_2O/kg to 0.72 mL/cm H_2O/kg. This may be because of the effect of high-frequency chest oscillation on mucociliary clearance.[16–17]

Limitations of Negative Pressure Ventilation

Chest cuirass NPV is associated with few complications; however, some limitations do exist. Attention must be paid to the skin where the chest cuirass is fitted. Correct placement of the chest cuirass is important to facilitate a good seal and to minimize skin injury. A relative contraindication to NPV is fixed upper airway obstruction, as negative pressure may exacerbate problems with air flow via the Bernoulli effect. As demonstrated in Case 2, this limitation of NPV may be avoided by the concurrent use of nasal continuous positive airway pressure. Another limitation is that there is not a simple method to measure minute ventilation. This lack of a monitoring capability may cause anxiety for the patient care team when NPV is first introduced and may partially explain the lack of widespread acceptance of NPV for acute respiratory failure.

Summary

In conclusion, the three patients presented help to demonstrate that noninvasive NPV may be used to suit different scenarios not previously reported. The effects of this relatively new mode of ventilation, especially with high-frequency external chest wall oscillation (secretion clearance mode), on mucociliary clearance are largely unexplored in humans. Clinical trials in conditions with severe airway plugging such as cystic fibrosis and acute chest syndrome in sickle cell disease would be highly informative. Additionally, there is a need for improved respiratory support for patients with immunosuppression. Chest cuirass NPV may provide a partial solution that avoids many of the pitfalls of previous ventilation strategies.

REFERENCES

1. Fulkerson WJ, MacIntyre NR. Complications of mechanical ventilation. Prob Respir Care 1991;4(1):1–135.
2. McCulloch TM, Bishop MJ. Complications of translaryngeal intubation (review). Clin Chest Med 1991;12(3):507–521.
3. de Blic J, Delacourt C, Scheinmann P. Ultrathin flexible bronchoscopy in neonatal intensive care units. Arch Dis Child 1991;66(12): 1383–1385.
4. Little LA, Koenig JC Jr, Newth CJL. Factors affecting accidental extubations in neonatal and pediatric intensive care patients. Crit Care Med 1990;18(2):163–165.
5. Dreyfuss D, Saumon G. Ventilator induced lung injury: lessons from experimental studies (review). Am J Respir Crit Care Med 1998; 157(1):294–323.
6. Kumar A, Pontoppidan H, Falke KJ, Wilson RS, Laver MB. Pulmonary barotrauma during mechanical ventilation. Crit Care Med 1973; 1(4):181–186.
7. Slavin G, Nunn JF, Crow J, Dore CJ. Bronchiolectasis: a complication of artificial ventilation. BMJ (Clin Res Ed) 1982;285(6346): 931–934.
8. Finlay-Jones JM, Papadimitriou JM, Barter RA. Pulmonary hyaline membrane: light and electron microscopic study of the early stage. J Pathol 1974;112(2):117–124.
9. Capers TH. Pulmonary hyaline membrane formation in the adult: a clinicopathologic study. Am J Med 1961;31(Nov):701–710.
10. Langer M, Mosconi P, Cigada M, Mandelli M. Long-term respiratory support and risk of pneumonia in critically ill patients. Intensive Care Unit Group of Infection Control. Am Rev Respir Dis 1989; 140(2):302–305.
11. Fagon JY, Chastre J, Domart Y, Trouillet JL, Pierre J, Darne C, Gibert C. Nosocomial pneumonia in patients receiving mechanical ventilation: prospective analysis of 52 episodes with use of a protected specimen brush and quantitative culture techniques. Am Rev Respir Dis 1989;139(4):877–884.
12. Fewel JE, Abendschein DR, Carlson CJ., Rapaport E, Murray JF. Mechanism of decreased right and left ventricular end-diastolic volumes during continuous positive-pressure ventilation in dogs. Circ Res 1980;47(3):467–472.
13. Tobias JD, Rasmussen GE. Pain management and sedation in the pediatric intensive care unit (review). Pediatr Clin North Am 1994; 41(6):1269–1292.
14. Thomson A. The role of negative pressure ventilation (review). Arch Dis Child 1997;77(5):454–458.
15. Petros AJ, Fernando SS, Shenoy VS, al-Saady NM. The Hayek Oscillator. Nomograms for tidal volume and minute ventilation using external high frequency oscillation. Anaesthesia 1995;50(7):601–606.
16. Gross D, Zidulka A, O'Brien C, Wight D, Fraser R, Rosenthal L, King M. Peripheral mucociliary clearance with high-frequency chest wall compression. J Appl Physiol 1985;58(4):1157–1163.
17. King M, Phillips DM, Gross D, Vartian V, Chang HK, Zidulka A. Enhanced tracheal mucus clearance with high frequency chest wall compression. Am Rev Respir Dis 1983;128(3):511–515.
18. Prabhakar G, Timberlake GA, Chendrasakhar A, Dulaney J, Reedy VS, Barringer EL. Effects of positive pressure and external chest wall oscillation on load independent cardiac function. Surgical Forum 1996;48:59–61.
19. Williams DB, Kiernan PD, Metke MP, Marsh HM, Danielson GK. Hemodynamic response to positive end-expiratory pressure following right atrium-pulmonary artery bypass (Fontan procedure). J Thorac Cardiovasc Surg 1984;87(6):856–861.
20. Shekerdemian LS, Bush A, Shore DF, Lincoln C, Redington AN. Cardiopulmonary interactions after Fontan operations: augmentation of cardiac output using negative pressure ventilation. Circulation 1997;96(11):3934–3942.
21. Shekerdemian LS, Shore DF, Lincoln C, Bush A, Redington AN. Negative-pressure ventilation improves cardiac output after right heart surgery. Circulation 1996;94(9 Suppl):II49-II55.
22. Murray JF, Felton CP, Garay SM, Goettlieb MS, Hopewll PC, Stover DE, Teirstein AS. Pulmonary complications of the acquired immunodeficiency syndrome: a report of a National Heart, Lung and Blood Institute workshop. N Eng J Med 1984;310(25):1682–1688.
23. Marolda J, Pace B, Bonforte RJ, Kotin N, Kattan M. Outcome of mechanical ventilation in children with acquired immunodeficiency syndrome. Pediatr Pulmonol 1989;7(4):230–234.
24. Wong HR, Chundu KR. Improved outcome for young children with AIDS, *Pneumocystis carinii* pneumonia, and acute respiratory failure. Pediatr Pulmonol 1994;18(2):114–118.
25. Sleasman JW, Hemenway C, Klien AS, Barrett DJ. Corticosteroids improve survival of children with AIDS and *Pneumocystis carinii* pneumonia. Am J Dis Child 1993;147(1):30–34.
26. Bye MR, Cairns-Bazarian AM, Ewig JM. Markedly reduced mortality associated with corticosteroid therapy of *Pneumocystis carinii* pneumonia in children with acquired immunodeficiency syndrome. Arch Pediatr Adolesc Med 1994;148(6)638–641.
27. Rivera R, Tibballs J. Complications of endotracheal intubation and mechanical ventilation in infants and children. Crit Care Med 1992; 20(2):193–199.
28. Kolobow T, Tsuno K, Extracorporeal membrane oxygenation and intravascular membrane oxygenation. In: Tobin M, editor. Principles and practice of mechanical ventilation. New York: McGraw Hill; 1994: 469.
29. Garg M, Lew CD, Ramos AD, Platzker AC, Keens TG. Serial measurement of pulmonary mechanics assists in weaning from extracorporeal membrane oxygenation in neonates with respiratory failure. Chest 1991;100(3):770–774.
30. Bonekat HW. Noninvasive ventilation in neuromuscular disease (review). Crit Care Clin 1998;14(4):775–797.
31. Chihara K, Ueno T, Itoi S, Nakai M, Sahara H, Oguri S, et al. Ventilatory support with a cuirass respirator after resection of bullous emphysema: report of a case. J Thorac Cardiovasc Surg 1996; 111(6):1281–1283.
32. Broomhead CJ, Dilkes MG, Monks PS. Use of the Hayek oscillator in a case of failed fibreoptic intubation. Br J Anaesth 1995;74(6):720–721.
33. Monks PS, Broomhead CJ, Dilkes MG, McKelvie P. The use of the Hayek Oscillator during microlaryngeal surgery. Anaesthesia 1995; 50(10):865–869.