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CASE REPORT

Combating Respiratory Failure in Guillain–Barré Syndrome within a Resource-Limited Rural ICU: A 7-Month Weaning Journey

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ABSTRACT

Background : Respiratory failure in Guillain–Barré syndrome (GBS) is the most dangerous complication. Approximately 22% of patients with GBS require a mechanical ventilation (MV) within the first week of hospital admission. Management of the patient with prolonged MV during ICU stay has been challenging, especially in resource-limited rural areas. The important consideration is the patient's ability to regain spontaneous breathing to evaluate the readiness to wean and the optimal time for deciding to decannulation.

Case Illustration : We reported a case about the management of respiratory failure due to GBS in the ICU involving a 37-year-old woman presented to the emergency room due to rapidly progressive weakness. The patient was treated with a 5-day course of intravenous immunoglobulin (IVIG). After 25 days with a MV, the patient underwent bedside PDT as a choice for critically ill patients who require prolonged MV in the ICU. The patient's motor development during the treatment period in the ICU showed a slow progression. There was no further significant progress in the patient's motor development for up to six months. Furthermore, on day 183 of the treatment, muscle recovery began to become obvious. We found a successful liberation of dependency after 207 days on MV and successful decannulation of the tracheostomy tube on day 215 in a hospital with limited facilities in rural areas. The patient was transferred from the ICU to the ward on day 217 and discharged on day 219. A one-month follow-up showed there were no respiratory complaints.

Conclusion: Long-term care for a GBS patient with prolonged MV in the ICU requires a multidisciplinary team approach, including optimization of treatment, nutrition, rehabilitation as well as psychological support. Comprehensive ICU care is the key to success in our case.

Keywords: Guillain–Barré syndrome, Mechanical ventilation, Percutaneous dilatational tracheostomy, Respiratory failure.

INTRODUCTION

GBS is a monophasic disease and a rare disease with an incidence of 0.81-1.89 per 100,000 people every year. GBS is a monophasic disease characterized by rapidly progressive bilateral extremity weakness combined with hyporeflexia or areflexia.[1] GBS patients will require ICU care if they experience respiratory distress, cardiovascular dysfunction (arrhythmia), severe swallowing disorders, reduced cough reflex, and rapid progression of weakness. As many as 22% of patients with GBS require a MV within the first week of hospital admission.[2] Respiratory failure is the most dangerous complication of GBS. The incidence of respiratory failure among adults with GBS varies between 6% to 33% across different countries, with up to 30% of patients requiring ventilatory support in the ICU.[3] Physicians must evaluate the patient for the possibility of a prolonged MV, moreover need to consider the benefits and risks of tracheostomy.[4] Percutaneous Dilatational Tracheostomy (PDT) is an increasingly developing technique considered as a choice for critically ill patients.[5,6,7] Management of GBS patients with prolonged MV will be challenging, especially in restoring respiratory function. Prolonged MV in GBS patients reached an average of 193 days. Weaning success reached 76%, while 31% of patients were successfully decannulated without respiratory support afterward.[8]

CASE ILLUSTRATION

A 37-year-old woman was presented to the emergency room due to her right leg suddenly could not be moved one day before admission. Progressive weakness started gradually from her right leg followed by her left leg and both arms. She had a tingling sensation in her mouth, headaches, and bloating in her belly. History of respiratory or gastrointestinal infections was denied. The patient's condition was compos mentis and moderately ill, with her vital signs showing blood pressure 140/90 mmHg, heart rate 115x/minute, respiratory rate 20x/minute, temperature 36 C, and SpO2 99% while breathing room air. Her body weight was 50 kg. On physical examination included head, thorax, and abdomen were within normal limits. Neurological examination showed negative for a meningeal sign, normal cranial nerves, motor strength was 221/122/000/122, no sensory

function deficit, no physiological reflexes included biceps, triceps, knee, and achilles, no pathological reflex deficit. Laboratory results showed Hb 12.7 g/dL, Ht 40.3%, Erythrocytes 5.65 million/microL, Leukocytes 11.2 thousand/mm3, Platelets 462 thousand/microL. Blood glucose was 105 mg/dL. Sodium 143 mmol/L, Potassium 3.4 mmol/L, Chloride 110 mmol/L. The initial diagnosis was acute tetraparesis due to suspected GBS and Myasthenia Gravis as a differential diagnosis. There were no bulbar symptoms, such as difficulty swallowing or slurred speech. MRC Sum Score at hospital admission was 15. Erasmus Guillain–Barré Syndrome Respiratory Insufficiency Score (EGRIS) was estimated 6. Initial treatment was given Methylprednisolone 62.5 mg BD IV and Omeprazole 40 mg BD IV.

On the second day of treatment, the patient suddenly had apnea and arrest in the ward. Cardiopulmonary resuscitation (CPR) was started for four cycles. The patient had a return of spontaneous circulation (ROSC). The patient was ventilated in a Pressure Controlled Ventilation (PCV) mode with Pressure 13 cmH2O, RR 20x/minute, PEEP 5 cmH2O, FiO2 60%. Vital signs post ROSC were normal. The patient was transferred to the ICU (MRC Sum Score was 0 and Hughes GBS disability scale was 5). On the 3rd day of treatment, the ventilator settings were changed to Synchronized Intermittent Mandatory Ventilation (SIMV) with Tidal Volume (TV) 6 ml/kg PBW, RR 16x/minute, PEEP 6 cmH2O, FiO2 60% with target SpO_2 more than 95%. Motor strength was 0/0/0/0. On the 4th day of treatment, the patient underwent a Prostigmin test using Neostigmin 1.5 mg and Sulfas atropine 0.5 mg. The patient showed no response. Therefore, the possibility of Myasthenia Gravis could be ruled out. In this case, three Brighton criteria were met, including bilateral flaccid weakness of the limbs, reduction or absence of deep tendon reflexes in the affected limbs, and a monophasic disease course with onset occurring between 12 hours to 28 days. However, a lumbar puncture and cerebrospinal fluid (CSF) analysis could not be performed due to limited resources.

On the 5th day of treatment, the patient was given IVIG. The first test dose vial was 0.01 ml/kg/minute for 30 minutes. There were no hypersensitivity

reactions or hemodynamic changes after the test dose, then IVIG administration was continued at a dose of 0.4 g/kg/day (20 g/day with body weight 50 kg) IV for five days. Total dose given was 100 g for 5 days without side effects. Lip movement was observed two weeks after IVIG administration.

After 25 days with a MV, there is still no significant improvement in the patient's breathing. The Hughes GBS disability scale was 5. Therefore, the patient was decided to undergo PDT due to the lack of respiratory muscle improvement and the risk of prolonged ventilation over 15 days. PDT was performed using a size 8.0 tube. The patient was positioned supine with the neck in hyperextension. The anatomical landmarks were identified between the second and third tracheal rings. Tracheal localization was achieved using a 10 mL syringe filled with lidocaine until the appearance of an air bubble. Subsequently, a 0.7 cm horizontal incision was made at the identified site to minimize the risk of bleeding. PDT was safely performed bedside in the ICU. On the 7th day of PDT treatment, the wound was kept clean and dry, also no signs of infection were found. Chest x-ray evaluation (Figure 1) showed the tracheostomy tube was located in the central hemithorax with a tip ± 2 cm from the carina. There is no visible sign of pneumonia on this chest x-ray. One week after PDT, the patient's risk of prolonged ventilation was assessed using a Spontaneous Breathing Trial (SBT), which demonstrated a tidal volume of less than 100 mL, indicating severe respiratory muscle weakness and a high risk of extubation failure. Negative inspiratory force (NIF) measurements were not performed due to unavailability of the equipment.

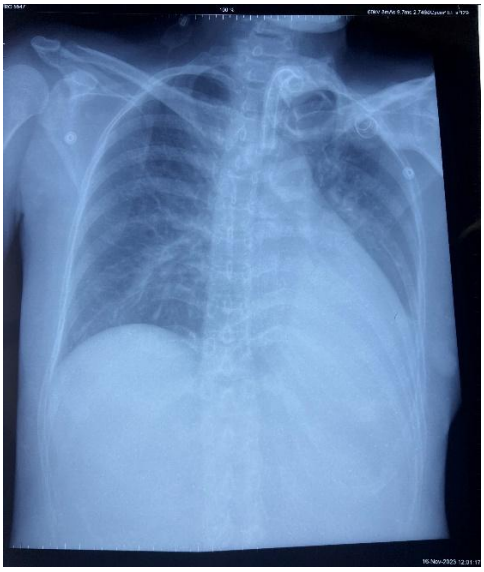


Figure 1. Chest x-ray evaluation post PDT

The patient's motor development during the treatment period in the ICU showed a slow progression which can be seen in Figure 2. On the 27th day post

IVIG, it was found that the patient was starting to be able to move the right shoulder, motor strength of the shoulder was 1/0. There was no further significant progress in the patient's motor development for up to six months. On day 183 of the treatment, muscle recovery began to become obvious. The patient was able to move her arms and legs with motor strength of 3/3/2/2. The MRC Sum Score was 28. During the treatment period, the patient experienced several recurrent complications such as ventilator-associated pneumonia (VAP), anemia, electrolyte disturbance, hypoalbuminemia, and urinary tract infection (UTI), which can be seen in Figures 3, 4, and 5. Prevention of complications that has been carried out in this case includes head of bed elevation 30-45^0, oral care, sedation vacation, subglottic suction, early mobilization, deep vein thrombosis prophylaxis, and bundle of urinary tract infection. The patient received comprehensive ICU care management according to the needs of the patient's condition. The patient underwent rehabilitation to improve the patient's well-being. The patient also received psychological support from family as well as physicians and nurses in the ICU. The nutritional targets for this patient were 25–30 kcal/kg/day with protein 1.2–2 g/kg/day including catabolic adjustments. The patient received enteral nutrition via a nasogastric tube (NGT). Two months following PDT placement, the NGT was removed. Aspiration was prevented by ensuring the patient had adequate swallowing and cough reflex.

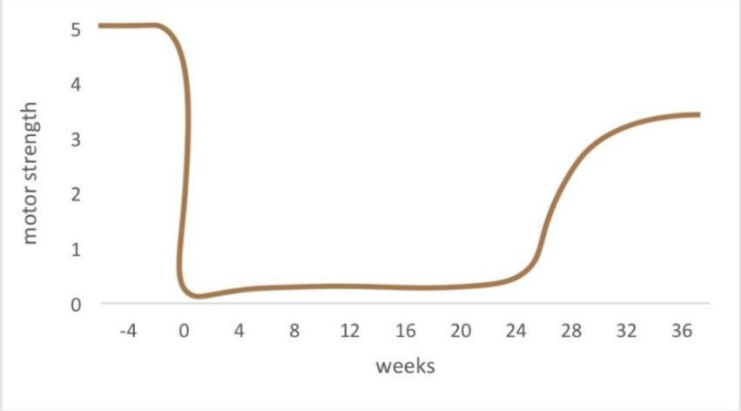


Figure 2. Progression of patient's motor development

On the 191st day, the patient began to experience adequate spontaneous breathing accompanied by regular and rhythmic chest movements with a stable hemodynamic condition. The Rapid Shallow Breathing Index (RSBI) was 53 breaths/minute/L (RR 16 x/minute, TV 300 mL), a score under 105 indicating a high likehood of successful extubation. There are several criteria for successful SBT in this patient including RR <35 breaths/ minute, HR <140 x/ minute, SpO2 >90% on FiO2 ≤40%, haemodynamic stability, no signs of increased work of breathing or distress. The patient also demonstrated an adequate ability to clear secretions and an effective cough, indicating sufficient airway protection. Weaning was initiated with the CPAP ventilator setting mode with PEEP 5 cmH_2O. The first SBT trial lasted only 10 minutes before it was terminated due to severe agitation and tachycardia (HR >140 x/ minute). The SBT was eventually successful, lasting for 120 minutes on the 16th day of weaning, after which extubation was performed. The patient was successfully weaned from MV after 207 days. Afterwards, the patient was using T-piece. The patient practiced coughing and speaking for seven days. We found that swallowing function was adequate, there was no dependency on MV, and the hemodynamic condition was stable. Finally, we decided to decannulate the tracheostomy tube on day 215. Post-decannulation, the patient was able to breathe spontaneously without any additional respiratory support. The patient was transferred from the ICU to the ward on day 217 and discharged on day 219. However, the patient was still unable to walk. The MRC Sum Score was 30 and Hughes GBS disability scale was 4. A one-month follow-up showed there were no respiratory complaints and the tracheostomy wound continued to heal well. However, the patient remained unable to perform activities of daily living independently.

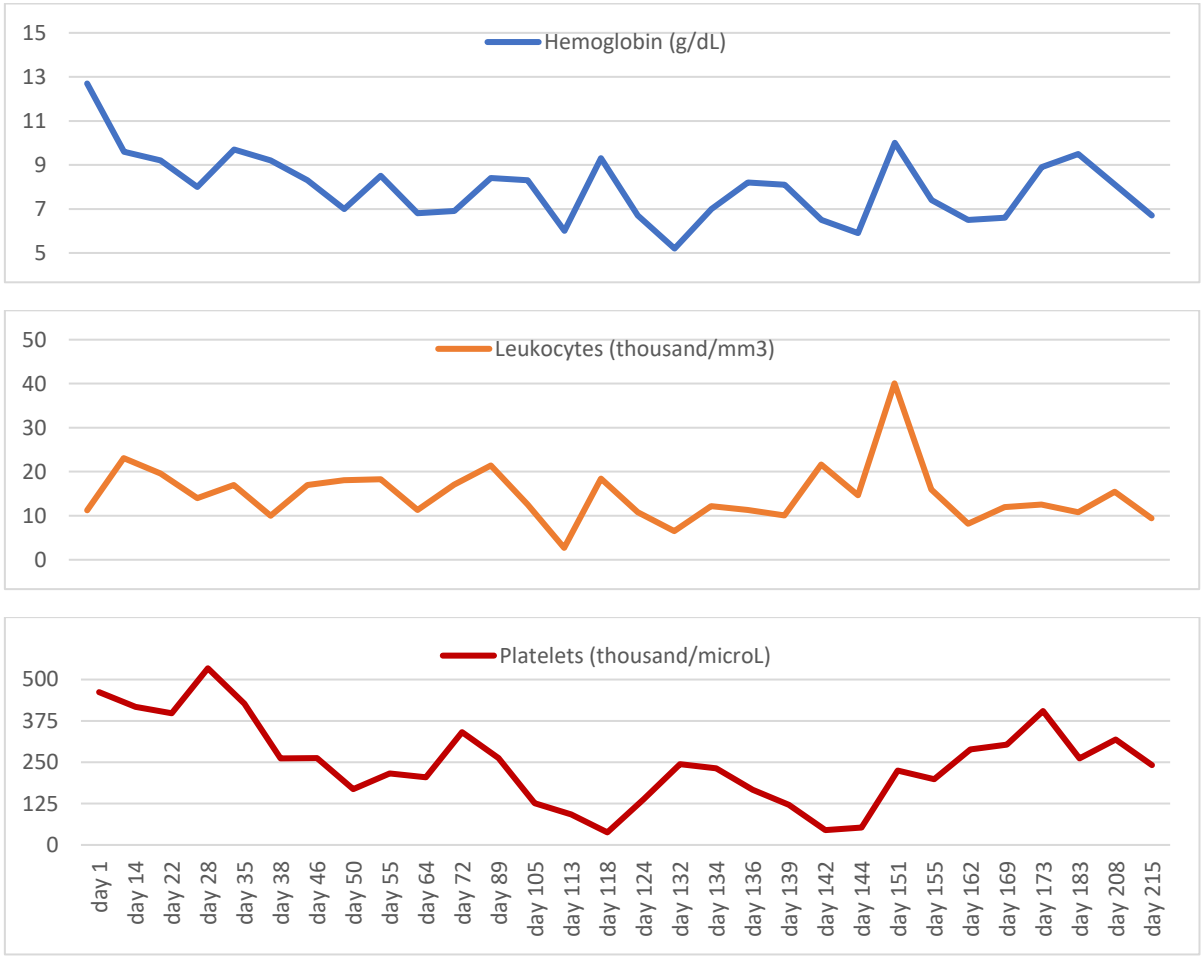
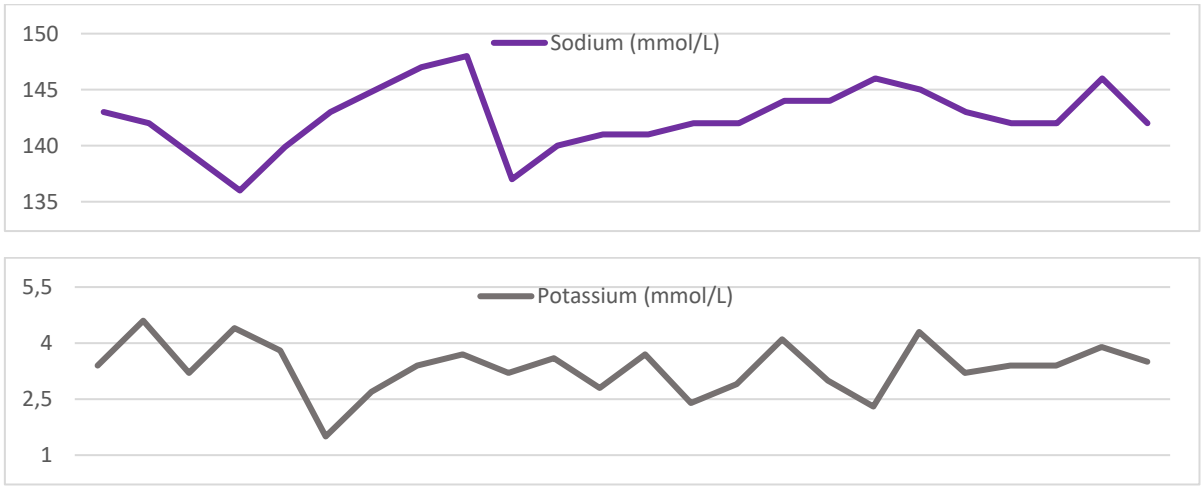


Figure 3. Trend of hemoglobin, leukocytes, platelets



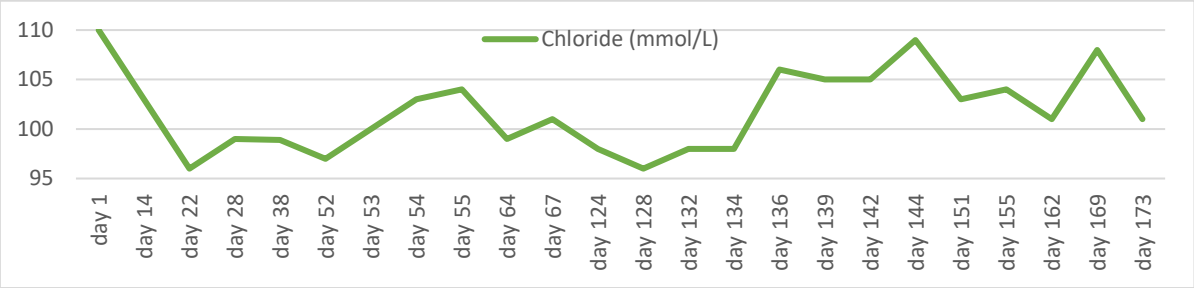


Figure 4. Trend of electrolytes

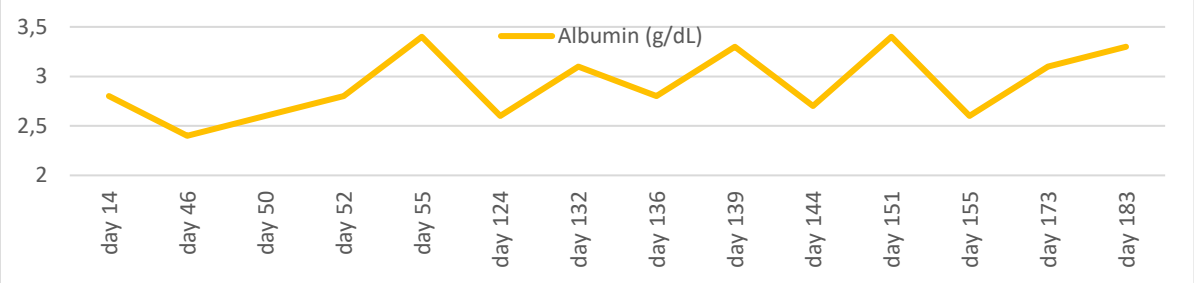


Figure 5. Trend of albumin

DISCUSSION

GBS is a rare disease with an incidence of 0.81-1.89 per 100,000 people every year. GBS occurs more often in men than women with a ratio of 3:2. The incidence will increase with age. Two-thirds of GBS patients generally experience respiratory or gastrointestinal infections before the onset of weakness. *Campylobacter jejuni* is the most common pathogen that causes infection. GBS is a monophasic disease that can be diagnosed clinically. GBS is characterized by rapidly progressive bilateral extremity weakness combined with hyporeflexia or areflexia. Weakness is ascending and generally starts from the distal part of the lower extremities. GBS can cause a variety of other symptoms including cranial nerve deficits, sensory symptoms, ataxia, pain, and autonomic dysfunction. The disease course of GBS generally reaches maximum severity (nadir) within four weeks. This progressive phase will be followed by a plateau phase for two days to six months before the patient begins to recover. GBS is classified into Acute inflammatory demyelinating polyneuropathy (AIDP), Acute motor axonal neuropathy (AMAN), Acute motor sensory axonal neuropathy (AMSAN), and Miller Fisher syndrome. Due to the absence of sufficiently sensitive and specific

biomarkers, the diagnosis of GBS is based on medical history and physical examination. In addition, the diagnosis can be supported by CSF and electrodiagnostic examinations. Complete blood counts, glucose, electrolytes, kidney function and liver enzymes are needed to rule out the other possible causes of acute flaccid paralysis including infection, metabolism, and electrolyte disorders. The classic finding in GBS patients through CSF examination is an increase in CSF protein levels and a normal CSF cell count, known as albumino-cytological dissociation. However, protein levels are generally normal in 30-50% of patients in the first week after onset and in 10-30% of patients in the second week. Therefore, a normal CSF protein level does not exclude the diagnosis of GBS. Electrodiagnostics with nerve conduction studies (NCS) is not necessary to diagnose GBS but can help confirm the diagnosis especially in atypical GBS. The characteristic of GBS is the discovery of a 'sural sparing pattern'.[1,2,9] In our case report, the diagnosis of GBS was based on clinical manifestations such as motor weakness in all extremities and areflexia. Laboratory examinations obtained normal results so that the other possible causes could be ruled out. Specific examinations for GBS are not carried out in our case due to limited facilities.

Management of GBS requires a combination of supportive treatment and immunotherapy. Effective treatments that have been proven for GBS are IVIg (0.4 gram/kg per day for 5 days) and plasma exchange (200-250 mL plasma/kg in 5 sessions). Both are equally effective treatments for GBS. The combination of treatment with plasma exchange followed by administration of IVIg did not show a significantly better effect than treatment with either plasma exchange or IVIg alone. Plasma exchange requires special equipment and is not always available in all hospitals, while IVIg is easier to administer and more widely available. Therefore, IVIg is used as the treatment option for GBS.[1,2] In severe cases, IVIg initiated within two weeks of disease onset showed a more rapid recovery effect. A systematic review and meta-analysis indicate that IVIG and PE have comparable therapeutic efficacy. Given its ease of administration, IVIG may therefore be considered the preferred

treatment option for GBS.[10,11] In our case, we use IVIg as the primary treatment because the availability of the drug is easier to obtain than plasma exchange.

Respiratory failure is the most dangerous complication of GBS which can occur suddenly. Progressive quadriplegia over 2 to 5 days can be a risk factor for the use of MV. Progressive respiratory muscle weakness involving inspiratory and expiratory muscles. Weakness of the diaphragm is caused by demyelination of the phrenic nerve. Vital capacity (VC) and total lung capacity (TLC) are reduced, residual volume (RV) is normal or increased, and RV/TLC ratio is high in the absence of airway obstruction. Paralysis of the abdominal and intercostal muscles will impair the ability to clear secretions by coughing. Facial and oropharyngeal muscle weakness will cause swallowing disorders which can lead to aspiration pneumonia. Bulbar dysfunction, bilateral facial weakness, and dysautonomia in GBS are closely related to the incidence of respiratory failure. All of these mechanisms will ultimately cause respiratory failure.[3,4] GBS patients will require ICU if they experience respiratory distress, cardiovascular dysfunction (arrhythmia), severe swallowing disorders, reduced cough reflex, and rapid progression of weakness. Approximately 22% of patients with GBS require a MV within the first week of hospital admission. Patients who are at risk of respiratory failure must be identified as early as possible. Erasmus GBS Respiratory Insufficiency Score (EGRIS) is a tool used to estimate the probability (1-90%) of determining a patient's need for a ventilator in the first week of hospital admission.[2] A measurement of vital capacity (VC) is the most frequently used method in clinical practice. It is simple to perform and noninvasive, although not feasible in acute settings. A reduction in VC of more than 20% when measured in the supine position indicates significant diaphragmatic weakness. Other bedside tests for assessing respiratory muscle strength include maximal inspiratory pressure (MIP) and maximal expiratory pressure (MEP). VC < 20 mL/kg, MIP > -30 cmH_2O, MEP < 40 cmH_2O is an important parameter to initiate MV in GBS. However, the initiation of MV is frequently determined through clinical judgment, as the presence of bulbar palsy may limit

the accuracy of functional measures such as VC, MIP, and MEP. In addition, diaphragmatic ultrasonography is a useful tool for assessing respiratory muscle function. The variation in diaphragmatic thickness during respiration can be quantified as the thickening fraction (TF). A TF of less than 20%, regardless of the presence of paradoxical diaphragmatic motion, indicates a need for immediate ventilatory support.[3]

Considerations in performing a tracheostomy in GBS patients are based on the long duration of intubation which can lead to the laryngeal nerve injury, laryngeal mucosa, vocal cord, and increase the risk of fistula formation.[4] Timing of tracheostomy is still controversial. The decision to tracheostomy is determined based on each patient individually, the causes of respiratory failure, the estimated duration with long-term MV, the number of weaning attempts, the risk of prolonged endotracheal tube use, and the risk of a tracheostomy. In clinical settings, indication and timing for tracheostomy is generally performed after 10 to 14 days of MV.[5] In GBS patients, reduced motor ability upon admission to the ICU or at the end of immunotherapy administration was significantly associated with a duration of prolonged MV for more than 15 days. Clinical observation after the patient is admitted to the hospital is carried out for at least 10-14 days before tracheostomy, which is expected to avoid unnecessary tracheostomy in GBS patients. The decision should be made as early as possible when rapid recovery cannot be achieved in GBS patients.[4] In our case, a tracheostomy was considered based on the clinical condition and the patient's response to the treatment, which has not shown any improvement, especially in the respiratory muscles. There was a risk of prolonged MV for more than 15 days. It was the first time a tracheostomy has been performed at this hospital in rural areas with elaborate equipment and team preparation.

Tracheostomy can be performed using a new technique described as PDT. Various studies showed that PDT is more beneficial in perioperative and postoperative complications compared with the surgical tracheostomy (ST). Other benefits are related to time duration and cost.[5,7] A study reported that PDT is an alternative to ST in ICU

patients, providing comparable outcomes with fewer complications including subcutaneous emphysema and postoperative bleeding. PDT is a safe technique to be performed bedside in the ICU, thereby eliminating the need to transport critically ill patients to the operating room, which is often difficult and increases the risk of complications. PDT is considered as a choice for critically ill patients who require a tracheostomy.[6] The ideal patient position is neck hyperextension. PDT is generally performed between the second and third tracheal rings.[5,12,13] A study showed that blind PDT and bronchoscopic guided PDT showed equivalent safety when blind PDT was performed by an experienced person.[14] In our case, PDT was chosen to be performed using an anatomical landmark approach technique without bronchoscopy guiding due to unavailability of the equipment.

Long-term treatment of GBS patients in the ICU can cause several complications, including ventilator-associated pneumonia (VAP)/ hospital-acquired pneumonia (HAP), neuropathic pain, deep venous thrombosis (DVT), pulmonary embolism, cardiovascular problems (arrhythmia), rhabdomyolysis, hyperglycemia, electrolyte disturbance, and psychological trauma. Complications that arise must be detected quickly and treated appropriately.[4] The ICU Liberation Bundle (ABCDEF Bundle) provides an integrated framework for optimizing critical care including assess, prevent, and manage pain (A), both spontaneous awakening and breathing trials (B), choice of sedation strategies (C), assess, prevent, and manage delirium (D), early mobility and exercise (E), and family engagement and empowerment (F). A study reported that implementation of the ICU Liberation Bundle in MV adult ICU patients was associated with significant reductions in the duration of mechanical ventilation and ICU length of stay (LOS).[15]

Management of GBS patients with prolonged MV will be challenging, especially in restoring respiratory function. The use of MV in GBS patients reached an average of 193 days. Weaning success reached 76%, while 31% of patients were successfully decannulated without respiratory support afterward.[8] Weaning in GBS patients is quite different. The time for weaning is a difficult consideration. Weaning must be

done gradually and extubation must be postponed until the respiratory muscles have completely recovered. The decision to wean cannot depend solely on evaluating limb muscle strength because diaphragm weakness may disappear before limb weakness.[4] A study showed that diaphragmatic strength increases during the weaning process in patients with GBS and myasthenia gravis.[3] A 30-minute SBT is considered sufficient to assess readiness for liberation from MV in the general ICU population. However, the applicability of this approach to patients requiring prolonged MV remains uncertain, as many have undergone tracheostomy and present with significant generalized weakness. To assess readiness for SBT in patient with prolonged MV, rapid shallow breathing index (RSBI), defined as the ratio of respiratory rate (breaths per minute) to tidal volume (liters), has been widely investigated.[16] There are still limited studies on weaning in GBS patients with prolonged MV. Pressure ventilation modes with a gradual reduction in inspiratory support should be preferred during weaning. Patients with GBS are particularly susceptible to atelectasis. Consequently, several centers use pressure ventilation during the day and volume-controlled ventilation at night. Successful extubation requires fulfillment of several criteria for neuromuscular patient, including an effective cough reflex, minimal airway secretions, absence of respiratory infection, and sufficient motor strength to lift the head. SBT may be initiated when the forced vital capacity exceeds 15 mL/kg and is recommended when the MIP exceeds −20 cmH_2O or the MEP exceeds +40 cmH_2O. In patients with severe GBS, the weaning process may last for several days to weeks. SBT is usually performed repeatedly, with the duration progressively increased as the patient's tolerance improves.[17] Tracheostomy decannulation can be considered if there is no dependency on MV for five days, stable hemodynamic, pCO2 <60 mmHg from the blood gas analysis, preserved cognition, adequate swallowing function, and no airway obstruction.[16] Prolonged MV in GBS is associated with a poor outcome, but patients can show persistent recovery progress for years.[18]

CONSLUSION

Long-term care for a GBS patient with prolonged MV in the ICU requires a multidisciplinary team approach, including optimization of treatment, nutrition, rehabilitation as well as psychological support. Comprehensive ICU care is the key to success in our case.

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