Positional Change Used to Manage Postextubation Respiratory Failure in a Child With Cerebral Palsy

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Dental treatment for patients with cerebral palsy (CP) is often performed under general anesthesia due to involuntary movements that can render dental treatment difficult. Since CP is often accompanied by spasticity, care must be taken when positioning patients during general anesthesia. We report the management of a 14-year-old girl with CP and epilepsy undergoing general anesthesia for dental treatment who experienced respiratory failure due to acute thoracoabdominal muscle hypertonia after extubation. She had a history of cardiac arrest due to respiratory failure caused by acute muscle hypertonia and successful resuscitation. General anesthesia was induced after careful positioning of the patient to prevent spastic muscle stretching, and the dental treatment was completed without complications. However, upon awakening after extubation, the patient developed respiratory failure due to acute muscle hypertonia. The patient was resedated and repositioned from a supine to a sitting position, and her symptoms improved. There was no recurrence of muscle hypertonia, and she recovered fully without complications. In this case, respiratory failure associated with acute muscle hypertonia was successfully managed by position change after initial treatment with positive-pressure ventilation and propofol. It is important to be prepared for the possibility of respiratory failure associated with acute muscle hypertonia and its countermeasures when providing general anesthesia for patients with CP.

Key Words: Cerebral palsy; Muscle hypertonia; General anesthesia; Respiratory failure; Repositioning.

Patients with cerebral palsy (CP) may have difficulty with dental treatment due to involuntary movements and uncontrolled muscle tone. In addition, patients with CP may also have intellectual disabilities/developmental disorders that can cause them to be less receptive to dental care. Therefore, treatment under general anesthesia is commonly performed for such patients. Patients with CP who have spasticity require careful positioning during general anesthesia to prevent dislocations and pressure sores. Particular attention should be paid to excessive stress on joints and muscles, which may exacerbate muscle tone.

We performed dental treatment under general anesthesia for a child with CP with spasticity. The patient progressed uneventfully from induction of general anesthesia to the end of treatment; however, acute thoracoabdominal muscle hypertonia occurred after extubation, which led to respiratory failure. We report a case in which the patient's signs and symptoms of respiratory failure improved after resedation and a positional change from the supine to the sitting position.

CASE PRESENTATION

A 14-year-old girl (height 116 cm; weight 18 kg; body mass index 13.4 kg/m^2) with CP, epilepsy, severe intellectual disabilities, and difficulty communicating presented for endodontic treatment of the left maxillary first molar secondary to dental caries. Since the patient was unable to cooperate, general anesthesia was planned. She had been taking valproic acid (300 mg/d) and lamotrigine (20 mg/d) for epilepsy and had been seizure free for more than 2 years. At 12 years of age, she experienced an episode of acute thoracoabdominal muscle hypertonia triggered by excessive straining while defecating that resulted in respiratory failure and

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cardiac arrest. She was successfully resuscitated by mask ventilation and chest compressions. Thereafter, botulinum therapy was started once every 4 months, after which the patient was free of further acute muscle hypertonic attacks.

Routine preoperative blood tests and a resting 12-lead electrocardiogram revealed no abnormalities. Preoperative chest radiographs showed no abnormalities except for mild scoliosis. During the preoperative physical examination, her mandible was noted to be normal size, maximum interincisal distance was approximately 40 mm, thyromental distance was approximately 70 mm, and head extension was adequate. No snoring or apnea was noted during sleep. However, the patient's Mallampati classification could not be evaluated due to poor cooperation.

No premedication was given on the day of treatment, and all daily medications were continued as usual. After entering the anesthesia room by her buggy wheelchair, the patient was transferred to the operating table and placed in a supine position. A cushion was placed under her knees and her posture adjusted so that her upper body was elevated several degrees to avoid hyperextension of her spastic muscles. General anesthesia was induced with sevoflurane (5%), oxygen (2 L/min), and nitrous oxide (4 L/min). The patient lost consciousness without restlessness or muscle hypertonia.

After establishing intravenous (IV) access and confirming ease of mask ventilation, an IV bolus of rocuronium (11 mg) was administered to facilitate tracheal intubation. A size 5, cuffed, standard oral endotracheal tube was used for nasotracheal intubation. Anesthesia was maintained with sevoflurane (2-3%), oxygen (1 L/min), air (2 L/min air), and a continuous infusion of propofol (50-67 μg/kg/min). A total of 3 mL of 2% lidocaine with 1:80 000 epinephrine (total: 60 mg lidocaine and 0.0375 mg epinephrine) was administered for local infiltration anesthesia. The patient's blood pressure (80-130/55-80 mm Hg), heart rate (100-130 beats/min), SpO_2 (98-99%), and end-expiratory CO_2 partial pressure (38-46 mm Hg) were well maintained. No apparent epileptic seizures or muscle hypertonia were observed intraoperatively.

After the dental treatment was completed, the headrest and backboard angles, which had been slightly modified during treatment, were returned to the same position as at the time of anesthesia induction, and all anesthetics were discontinued. She was extubated before awakening after confirming adequate respiratory function and minute ventilation.

The patient's respiratory status remained stable for several minutes after extubation, but muscle thoracoabdominal hypertonia occurred upon awakening and resulted in respiratory failure. At that time, no spasms or muscle contractures in the limbs were observed. Her SpO_2 temporarily dropped to 82% but quickly improved to 99% with supplemental oxygen (6 L/min) via mask-assisted ventilation. There was no coughing or wheezing during this time. The muscle hypertonia persisted as did the respiratory failure without assisted ventilation. She did not lose consciousness during this period and had an anguished expression due to the ongoing respiratory distress. After administering a small bolus of propofol (20 mg), the patient lost consciousness, and the muscle hypertonia disappeared. However, the patient again presented with unstable breathing upon awakening.

Her mother had previously told us that the muscle hypertonia tended to subside when she was placed in a sitting and mildly forward-bent posture. Therefore, we administered another bolus of propofol (20 mg), transferred her to the buggy wheelchair, and postured her in a sitting position while sedated, carefully monitoring her respiratory status throughout. Airway patency was well maintained with stable spontaneous breathing. Later, the patient emerged from anesthesia and exhibited purposeful body movements with no further recurrence of the acute muscle hypertonia. After confirming that her SpO_2 was maintained above 97% under room air conditions, the patient was transferred to the recovery room. The operative time was 2 hours 57 minutes, the anesthesia time was 4 hours 12 minutes, and the infusion volume was 277 mL. She recovered from anesthesia with a stable respiratory status and no recurrence of muscle hypertonia. After confirming that the patient was fully awake and back to baseline consciousness, she was discharged home approximately 3 hours after the end of anesthesia. She had an uneventful course after returning home and continued to receive regular dental care at our clinic.

DISCUSSION

CP encompasses a group of neurologic disorders that permanently affect body movements, posture, and muscle coordination due to nonprogressive lesions attributed to abnormalities in or damage to the brain, typically occurring between conception and 4 weeks of birth. The frequency of CP has been reported to be approximately 2 per 1000 live births.[1] Patients may have difficulty undergoing dental treatment due to involuntary movements, muscle spasticity, and intellectual disabilities/developmental disorders. Therefore, it is common for patients with CP to be treated under IV sedation or general anesthesia.

Among the complications of CP, respiratory disorders are particularly problematic during anesthetic manage-

ment. These may include retention of airway secretions, upper airway or tracheal stenosis, scoliosis, central apneic attacks, and respiratory motor dysfunction.[2,3] Respiratory motor dysfunction is caused by a breakdown in coordination between the muscle groups involved in inspiration and expiration. Efficient spontaneous ventilation is achieved when the expiratory and inspiratory muscles relax and contract in a coordinated manner. In patients with CP, this muscle coordination may be impaired. The resulting respiratory failure can lead to severe hypoxemia, which may be fatal. The patient in our case had a history of uncontrolled muscle hypertonia triggered by defecation, which led to respiratory failure and cardiac arrest. Muscle hypertonia and respiratory failure occurred again after extubation once the patient awoke from anesthesia. Extubation can cause significant discomfort in the pharyngeal and laryngeal regions and trigger increased muscle tension due to coughing. This discomfort may have triggered muscle hypertonia, leading to respiratory failure. During the attack, the mental anguish and excessive muscle tension caused by respiratory distress further impaired respiratory motor coordination, resulting in a vicious cycle.

$GABA_A$-positive allosteric modulators act to improve muscle spasticity.[4] In this case, propofol, a sedative and positive allosteric modulator of $GABA_A$, was administered during an acute muscle hypertonia episode and produced rapid improvement. Treatment for muscle hypertonia associated with CP includes neurosurgery, intrathecal baclofen therapy, and botulinum toxin therapy.[5] Botulinum therapy was introduced to the patient after the first attack, and her muscle spasticity symptoms improved. The effect of botulinum injection lasts ~3 to 4 months but diminishes over time.[6] In this case, on the day of treatment, more than 2 months had passed since the patient had received her last botulinum injection. It may have been more ideal for general anesthesia to be scheduled closer to the date of her last botulinum injection to reduce the risk of another muscle hypertonia episode.

It is important to note that muscle hypertonia attacks and epileptic tonic-clonic seizures may be similar in appearance. Approximately 1 in 4 patients with CP are reported to have epilepsy, which was also present in our patient's case.[7] Although distinguishing between the two may be difficult, tonic-clonic seizures are associated with loss of consciousness.[8] In the present case, it was observed that the patient remained conscious during the attack, and it was determined that the patient was likely suffering from muscle hypertonia instead.

It is also important to differentiate muscle hypertonia from airway obstruction due to laryngospasm. If patients are spontaneously ventilating, clinical features of laryngospasm commonly include wheezing upon inspiration and the presence of rocking-type movements of the chest wall and abdomen given that the respiratory drive is otherwise intact. None of these signs were noted despite the patient spontaneously ventilating beforehand, and the patient had persistent thoracoabdominal muscle tone that we attributed to respiratory failure associated with muscle hypertonia. Again, differentiating between these complications may be difficult. However, in either case, positive-pressure ventilation (PPV) with jaw thrust maneuver and administration of propofol, as was done in this case, is often effective. In cases of sustained, complete laryngospasm, rapid paralysis with a neuromuscular blocking agent (ie, succinylcholine) may also be necessary.[9]

Respiratory function associated with CP can also be affected by body position. Studies examining respiratory function and posture changes in children with CP have reported that the supine position was most unfavorable compared with the sitting or side-lying positions.[10,11] In addition, the supine position is more likely to induce muscle hypertonia due to strain on spastic joints and muscle overextension. On the other hand, anesthesia induction, dental treatment, and extubation are generally performed with the patient placed in the supine position. In this case, the patient was placed in a supine position before induction of anesthesia, and then to avoid hyperextension of spastic muscles, we placed a cushion under the knees and placed the patient in a slightly elevated upper body position. However, muscle hypertonia still occurred after extubation.

We observed improvement after transferring the patient to the buggy wheelchair and positioning her in a sitting and slightly forward-bent position while the attack persisted. The buggy wheelchair, which the patient rides daily, facilitates stable positions that relieve muscle tension. On the other hand, being away from the operating table in a sitting position while recovering from anesthesia may delay effective emergency responses. The reclining seat function of her buggy wheelchair allowed immediate repositioning to the supine position, and we felt it possible to respond immediately if assisted ventilation was required. As a precautionary measure against the recurrence of muscle hypertonia after awakening, we decided to monitor the patient's recovery from anesthesia with the patient sitting in the buggy wheelchair while carefully observing the patient's respiratory status by monitoring with pulse oximetry and observing her abdominothoracic movements.

CONCLUSION

We experienced a case of respiratory failure attributed to acute muscle hypertonia after extubation for a patient

with CP who underwent general anesthesia for dental treatment. We first managed the patient with PPV and small boluses of propofol and then repositioned her in the sitting position in her buggy wheelchair. This strategy was successful in managing the patient without a recurrence during recovery. Despite its common use during induction, extubation, and dental care, the supine position may be detrimental to respiratory function in some patients with CP. It may also be valuable to determine preoperatively stable positions for patients that may help relieve muscle tension when managing acute muscle hypertonia attacks.

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