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Cricothyrotomy for an Unexpected Cannot Intubate, Cannot Ventilate Situation for a Patient with Chronic Graft-Versus-Host Disease After Induction of General Anesthesia: A Case Report

Authors' Contribution:

Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Patient: Male, 45-year-old
Final Diagnosis: Cannot intubate cannot ventilate
Symptoms: Airway obstruction
Clinical Procedure: —
Specialty: Anesthesiology

Objective: Unusual clinical course

Background: Chronic graft-versus-host disease (GVHD) is a major complication of hematopoietic stem cell transplantations. Due to fibrotic changes, patients with GVHD are at risk for difficult airway management. We encountered a case of chronic GVHD that went into a “cannot intubate, cannot ventilate” (CICV) condition after induction of general anesthesia and was managed using cricothyrotomy.

Case Report: A 45-year-old man with uncontrolled chronic GVHD developed pneumothorax of the right lung. Thoracoscopic dissection of the adhesions, closure of the pneumostomy, and drainage under general anesthesia were planned. In the preoperative airway assessment, we concluded that using a video laryngoscope or endotracheal fiber would be sufficient to intubate the patient after sedation and that airway management after the loss of consciousness would not be difficult. Therefore, general anesthesia was induced by rapid induction; however, the patient developed difficult mask ventilation. Intubation was attempted via a video laryngoscope or broncho-fiber but failed. Ventilating using a supraglottic instrument was difficult. The patient was evaluated to have a CICV condition. Thereafter, because of a rapid decrease in oxygen saturation (SpO₂) and bradycardia, a cricothyrotomy was performed. Subsequently, ventilation became adequate, SpO₂ increased immediately and drastically, and respiration and circulatory dynamics recovered.

Conclusions: We believe that anesthesiologists should practice, prepare, and simulate airway emergencies that can be experienced during surgery. In this case, we recognized that skin sclerosis in the neck and chest could lead to CICV. It may be suitable for airway management of scleroderma-like patients to select conscious intubation with a bronchoscope as a first choice.


Keywords: Airway Management • Graft vs Host Disease • Tracheotomy • Anesthesia

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Background

Chronic graft-versus-host disease (GVHD) is a major complication of hematopoietic stem cell transplantation and is caused by an immune response from transplanted T cells from the donor [1]. Chronic GVHD, which occurs due to an inflammatory process involving tissue damage and repair, has various clinical features that resemble autoimmune diseases, such as scleroderma [2-5]. Patients with scleroderma-like systemic sclerotic lesions can require bronchoscopy for the management of mask ventilation difficulties and airway clearance for a small mouth opening and inadequate neck extension owing to fibrotic changes in the temporomandibular joint and neck [6]. Failure of airway management during anesthesia induction is one of the leading causes of cardiac arrest and death. It is critical for anesthesiologists to maintain a patent airway during anesthesia induction. Currently, guidelines and algorithms for difficult airway management are being disseminated [7,8]. These guidelines elucidate appropriate responses to airway emergencies and identify the frequency and risk factors of various airway clearance difficulties [9]. Despite these measures, “cannot intubate, cannot ventilate” (CICV) situations can occur in 0.02% to 0.4% of cases; however, no systematic means of accurately predicting airway security and mask ventilation difficulties have been established [9-11].

Emergency surgical airway (ESA) is the final management option for CICV situations in difficult airway management cases; however, this procedure is rarely practiced and might be difficult in high-risk emergency cases [9,10,12]. Although perioperative management of chronic GVHD requires individualized management [13], cases of patients with GVHD who develop CICV conditions and high-risk airway emergencies during anesthesia induction have not been reported.

Herein, we describe a patient with sclerosing skin, fascia, and joint lesions having chronic GVHD, who developed a CICV condition after induction of general anesthesia and recovered after a cricothyrotomy was performed by an anesthesiologist.

Case Report

A 45-year-old man (height: 169 cm; weight: 44 kg) underwent chemotherapy and allogeneic bone marrow transplantation for acute lymphoblastic leukemia 4 years previously. The patient had no general anesthesia history and anatomical abnormalities before bone marrow transplantation. He maintained complete remission; however, chronic GVHD was difficult to control. Two years later, he developed pneumothorax in the right lung, associated with chronic GVHD. Thoracoscopic dissection of adhesions, closure of the pneumostomy, and drainage under general anesthesia was planned.

In the preoperative airway assessment, a Mallampati classification score III was obtained, with an upper lip bite test score of grade II and a slight limitation of forward mandibular movement. Moreover, he had sclerotic lesions in the body caused by chronic GVHD, resulting in a limited range of motion of the neck. Despite a mild limitation of mouth opening, at least 2 figure lengths were intact. Obesity and preoperative sleep apnea were not observed. Pulmonary function showed restrictive ventilatory impairment, with a vital capacity of 48.2% and forced expiratory volume of 1.0% to 80%.

Several experienced anesthesiologists discussed strategies for airway management before the induction of general anesthesia. To confirm whether mask ventilation was possible, we fitted the patient with a mask while he was awake. The patient had no facial deformities and had a good mask fit, which resulted in good face mask ventilation under spontaneous breathing. Furthermore, the patient had no mustache or missing teeth; therefore, mask ventilation was not difficult. The patient had cervical stiffness and some mouth opening limitation; however, no shortening of the distance between the chin and hyoid bone, obesity, or signs of upper airway stenosis were observed. Therefore, we concluded that using a video laryngoscope or endotracheal fiber would be sufficient to intubate the patient, and that airway management after the loss of consciousness would not likely be difficult. In addition, awake intubation with a double-lumen tube is associated with patient distress. Based on these points, we selected general anesthesia via rapid induction rather than conscious intubation.

The patient was pre-oxygenated with 6 L/min of oxygen before the induction of general anesthesia. Rapid induction was performed using 50 mg propofol, 50 mg rocuronium, and 50 µg fentanyl. After the loss of consciousness, the patient experienced difficult mask ventilation. Intubation was planned with a 37-Fr double-lumen tube using a video laryngoscope (McGrath MAC Model AO3, USA). However, upon insertion of the video laryngoscope into the patient's mouth with an attempt to expand the field of view, the vocal cords and epiglottis could not be observed. Lifting of the larynx was attempted to expand it; however, this was not possible due to cervical skin sclerosis. We promptly switched to intubation using a normal 7.5-mm cuffed tracheal tube to secure the airway. Another anesthesiologist attempted intubation and similarly found a grade 4 on the Cormack grading scale. In accordance with the Japanese Society of Anesthesiologists–American Medical Association (JSA-AMA) guidelines, we attempted to intubate the patient using a method that could be performed immediately [8]. Bronchoscopic intubation was then attempted. The middle pharynx was observed, and no sclerosis in the surrounding tissues was found. Glossoptosis prevented visualization below the lower pharynx, the epiglottis and glottis were completely obscured, and the bronchoscope could not be guided

into the trachea. Even if the caregiver raised the mandible, it could not be lifted owing to skin contracture, and a view of the pharynx could not be established. In the next step, a supraglottic instrument (i-gel #4, Intersurgical, UK) was inserted; air leakage was strong, and ventilation was not possible. At this point, the patient was evaluated to be in a CICV situation; in the yellow zone, consciousness was restored and he resumed spontaneous breathing. Thereafter, with a rapid decrease in oxygen saturation (SpO₂) from 96% to 30%, the patient gradually became bradycardic, and we determined that the patient had entered the red zone [8]. Therefore, a cricothyrotomy was performed. After incising the skin and cricothyroid ligament using a scalpel, the incision was dilated using a pean. Finally, a 7.5-mm cuffed tracheal tube was inserted using a rhinoscope to expand the incision further and establish the endotracheal view.

After intubation, the patient was well ventilated; SpO₂ immediately and drastically increased, heart rate recovered, and respiratory and circulatory dynamics stabilized before any adverse events due to the CICV condition occurred. Surgery for pneumothorax was thereby cancelled. Emergency tracheostomy was performed immediately by an otorhinolaryngologist, who closed the cricothyroid ligament incision. The patient was admitted to the intensive care unit (ICU), without a cardiovascular agonist infusion. Soon after admission, his level of consciousness recovered to a Full Outline of UnResponsiveness score of 14 (4-4-4-2). The patient was discharged from the ICU on postoperative day (POD) 2 without apparent postoperative complications. Fortunately, the pneumothorax healed as a result of continued conservative treatment, and the chest drain was removed on POD 10. The treatment needed a prolonged period of time because of postoperative aspiration pneumonia in addition to the inherent poor respiratory condition due to restricting ventilation failure. Given the possibility of respiratory failure and re-intubation, we could not close the tracheostomy hole until the condition was stabilized. On POD 66, we were able to close the tracheostomy hole. On POD 84, the patient was transferred to another hospital for rehabilitation.

Discussion

We encountered a case of a CICV condition after general anesthesia induction in a patient with chronic cutaneous GVHD, who progressed into the red zone but eventually responded to management according to the JSA-AMA guidelines. ESA was performed as a life-saving measure. Although the number of fatal complications of general anesthesia is decreasing annually, the number of anesthesia-related severe hypoxic events per 10 000 anesthesia cases was 0.59 to 0.78 in Japan [14]. Anesthesiologists should prepare for and respond calmly to these rare emergencies.

The incidence of difficult airway management has also been reported previously. Shiga et al reported that the incidence of difficult intubation was 5.8% among 50 760 patients in 35 studies [10]. Nagaro et al reported that the incidence of CICV is 0.02% [11]. However, in a study of 4 tertiary care hospitals, 698 of 176 679 patients (0.40%) were found to have both mask and intubation insufficiencies [9]. Although there are some variations, CICV is rare. Independent predictors of difficult mask ventilation include cervical radiation dose changes, male sex, sleep apnea, Mallampati class III or IV, and having beards [15]. In this case, the male sex and Mallampati class III or IV were relevant (hazard ratio 5.8 [3.6-9.2]); however, the probability of predicting difficult mask ventilation was low.

Physical examination findings that best predict difficult intubation include an upper lip bite test score of grade III (lower incisors cannot extend to reach the upper lip), shorter horizontal distance, retrognathia (mandible measuring <9 cm from the angle of the jaw to the tip of the chin or subjectively short), and the widely used modified Mallampati score (≥ 3); however, none of these findings were applicable in our case [16].

In contrast, according to the difficult mask ventilation combined with difficult laryngoscopy prediction score, this case was risk class III (5 risk factors: Mallampati class III, male sex, presence of teeth, limited neck extension, and limited jaw protrusion), which suggested that the possibility of a CICV situation was 0.77% [9]. In conclusion, the preoperative airway assessment in this case was not positively predictive of intubation and mask ventilation difficulties, although it cannot be ruled out.

Reports on anesthetic cases in patients with chronic cutaneous GVHD are limited. In the present case, sclerodermitis and joint contractures were observed in the neck and thorax, and the symptoms were similar to those of scleroderma. Mask ventilation and intubation difficulties have been reported to occur due to microstomia and poor cervical extension because of fibrosis of the temporomandibular joint and neck caused by scleroderma [6,17]. Carr et al also provided a limited overview of airway management for scleroderma complications reported in 1990. Of the 8 patients with scleroderma in 7 studies, 1 patient was operated on with moderate sedation using dexmedetomidine and ketamine without intubation, 1 had unexpected airway compromise and was intubated using transtracheal wire guidance, 2 underwent awake fiberoptic intubation, 3 were switched to spinal anesthesia, 1 was managed using a Shikani intubating stylet, and 1 was intubated using a direct laryngoscopy [6]. It has been demonstrated that conscious intubation is not always performed; however, CICV has not been reported.

The reason why the patient in this case had a CICV situation despite the use of all available devices and why the supraglottic

devices were ineffective is unclear. Moreover, we wondered why the CICV situation could not be predicted. Verification should have been conducted.

Comparing this case with previous cases of CICV, we believe that skin sclerosis is a relevant risk factor, which was beyond our expectations. The patient had difficult mask ventilation soon after the induction of anesthesia, which was probably due to tongue root depression. Anatomically, the airway is opened by the elevation of the mandible by the anesthesiologist; however, in this case, the mandible failed to move due to cervical skin stiffness. For the same reason, we failed to expand the field of view of the pharynx using the video laryngoscope. Furthermore, because the mandible could not be raised, the bronchoscope could not be guided into the trachea because the field of view was obstructed by the sinking of the root of the tongue. Poor ventilation continued even after the insertion of the supraglottic device.

Gordon et al reported factors of difficulty with supraglottic airway device insertion and ventilation [18]. According to their report, fixation of the cervical spine is included; however, cervical skin sclerosis is not a risk factor. Additionally, chest wall abnormalities are a risk factor for difficulty in supraglottic device ventilation. In the present case, the patient had a restrictive ventilation problem due to skin contraction of the chest wall, which may have affected airway clearance with supraglottic devices.

Based on these findings, the most likely cause of airway clearance and mask ventilation difficulties in this clinical case was the skin stiffness from the neck to the chest. We believe that sclerotic skin lesions may be considered a new and extremely important risk factor for the CICV situation. Regardless of

the inspection of the difficult airway or the risk indicated by several guidelines, in a patient with scleroderma-like, it is always prudent and advisable to carry out conscious intubation with a bronchoscope as a first choice in order to avoid the ESA situation.

It has been reported that the causes of ESA include poor planning, poor judgment, deviation from recognized algorithms, and failure of technical skills [10]; however, we planned the anesthesia-induction method several weeks in advance and performed the positional test the day before. The consensus of some skilled anesthesiologists was that conscious intubation should have been selected. Nevertheless, we followed the guidelines for difficult airway clearance and responded quickly. However, because the patient had severe restrictive airway compromise and his oxygenation reserve was low, the oxygen saturation rapidly decreased and the patient fell into the red zone, leading to ESA. Although it was unclear whether awake intubation would have been successful in this patient, we concluded that the possibility of CICV could not be predicted in our case. In each case, we believe that anesthesiologists should practice, prepare, and simulate airway emergencies that can be experienced during the surgery.

Conclusions

We reported the general anesthesia management of a patient with chronic cutaneous GVHD and generalized sclerotic lesions who developed a CICV condition after general anesthesia induction. In this clinical case, we recognized that skin sclerosis in the neck and chest could lead to CICV. Conscious intubation with a bronchoscope should be considered as a first choice in the airway management of scleroderma-like patients.

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