Airway Surgery in Tracheostomised Patients with Wegener Granulomatosis Leading to Subglottic Stenosis

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Wegener granulomatosis (WG) is a multisystemic disorder characterised by granulomatous inflammation of the respiratory system. The growing of proliferative tissue towards the larynx and trachea may cause airway obstruction on account of subglottic stenosis. In this situation, the surgical goal is to eliminate the airway obstruction by providing natural airway anatomy. While mild lesions do not require surgical intervention, in fixed lesions, surgical intervention is required, such as tracheostomy, laser resection and dilatation. In tracheostomised patients, granuloma formation surrounding the tracheostomy cannula may occur in the trachea. Inflammation and newly formed granulation tissue result in severe stenosis in the airways. During surgical treatment of such patients, airway management is important. In this case report, we will discuss gas exchange and airway management with jet ventilation (JV) during excision of the granulation tissue with endolaryngeal laser surgery, leading to subglottic stenosis in tracheostomised patients in WG.

Keywords: Endolaryngeal laser surgery, jet ventilation (JV), subglottic stenosis, Wegener granulomatosis

Introduction

Wegener's granulomatosis (WG) is a multisystemic disease progressing with granulomatous inflammation of the respiratory system and characterised by necrotising vasculitis and granuloma formation, and its tracheobronchial involvements lead to stenosis (1). Subglottic stenosis found in 10–20% of WG patients can develop as the first finding of the disease or during the course of the disease (2). In this case, the surgical target is to eliminate the airway obstruction by providing a natural airway anatomy. While mild cases do not require surgical intervention, more severe fixed lesions require surgical interventions such as tracheostomy, laser resection and dilatation.

In patients who underwent tracheostomy, granulomas can occur in the tracheal tissue surrounding the tracheostomy cannula. Endotracheal or tracheostomy tube cuffs lead to pressure on airways and mucosal trauma, and this excessive pressure causes the formation of a necrotic area as a circle, leading to eschar and stenosis (3). Moreover, the development of inflammation and granulation tissue can form a serious stenosis in the airway. As a result, after opening tracheostomy, decannulation of these patients can be difficult due to continuous progression of the disease and restenosis. During the surgical procedures of these patients, airway control is very important. If surgical intervention is for airway reconstruction, the situation can be more difficult and more complicated. In this case report, gas exchange and airway management provided by jet ventilation (JV) during the excision of granulation tissue, which causes subglottic stenosis in patients with WG, using endolaryngeal laser surgery is discussed.

Case Presentations

Case 1

A 43-year-old patient who was receiving medical treatment after the diagnosis of WG for 4 years was opened tracheostomy 6 months ago because of subglottic stenosis associated with his disease. The patient, who had a complaint of difficulty in breathing from 3 months after the tracheostomy, planned to undergo tracheobronchoscopic examination under general anaesthesia and was then taken to the operating room. Following standard monitoring, a vascular access was established, and a fluid was given to the patient whose written informed consent was previously obtained. The patient weighed 86 kg, and anaesthesia induction was provided with 2 mg of midazolam, 100 µg kg⁻¹ of fentanyl, 200 mg of propofol and 16 mg of

mivacurium. After performing laryngoscopy with Macintosh blade (size 3), tracheoscopy was performed using a 4-mm diameter endoscope including a 0° optic (Karl Storz GmbH&Co, Tuttlingen, Germany). The obtained image revealed a granulation tissue surrounding the tracheostomy cannula at the subglottic level, causing stenosis in the trachea (Figure 1).

Thereupon, the excision of the granulation tissue with CO₂ laser microsurgery under general anaesthesia was planned for the patient. However, after removing the tracheostomy cannula, it was observed that the tracheal lumen was too narrow (4.3 mm), and it was impossible to perform surgical intervention in the presence of any tube. Then, a laser-resistant JV catheter with an external diameter of 3.4 mm and a length of 40 cm (Acutronic Medical Systems AG, Hirzel, Switzerland) was inserted. The parameters of JV were regulated in a way that the driving pressure (DP) was 1.2 bar, inspiration time (IT) was 50%, FiO₂ was 0.8, frequency was 130 and humidification level was 6. In addition to standard monitoring, ET-CO₂ was intermittently monitored with the help of a gas analyser in the JV device.

The maintenance of anaesthesia was provided with propofol at the rate of 6–10 mg kg⁻¹ h⁻¹ and remifentanyl at a rate of 0.05–0.25 µg kg⁻¹ min⁻¹. Moreover, 8 mg of dexamethasone, 50 mg of ranitidine, 10 mg of metoclopramide and 1 mg kg⁻¹ of prednisolone were intraoperatively administered.

One minute before the use of laser, JV settings were changed, and the pre-set laser mode was activated as FiO₂ of 0.30. In the 30th minute of the 95-min process, when SpO₂ decreased to 92–93%, DP, IT and FiO₂ increased to 1.3 bar, 60% and 0.35, respectively. Adequate oxygenation was enabled with JV at these settings, and the procedure was completed with the saturation level of 97–98% without any decrease (Table 1). An uneventful surgical vision was provided owing to the fine JV catheter, and the granulation tissue was resected with the CO₂ laser (Figure 2). With the CO₂ laser switched off, the laser mode of JV was closed, and the initial settings were returned. For eliminating CO₂, the frequency was adjusted to 300 and DP was adjusted to 0.8 for 1 min. Once spontaneous respiratory effort began in 2–3 min, the JV catheter was removed, and respiratory support was provided with a mask. In the patient observed to have sufficient spontaneous respiration, the insertion of tracheostomy cannula was not needed. The patient was decannulated and taken into the recovery room. Because respiratory distress did not develop in the recovery room, the patient was transferred to the clinic.

Case 2

A 59-year-old female patient who was followed-up due to WG for 1 year applied to the outpatient clinic of otorhinolaryngology with the complaint of respiratory distress. In her CT, stenosis narrowing the lumen in the subglottic area was revealed, and therefore, tracheostomy was opened. Two months after this procedure, the patient applied again due to recurring respiratory complaints and tracheoscopy was planned.

Figure 1. Tracheal stenosis

Table 1. Intraoperative haemodynamic parameters of the patients

		PHR (beat min ⁻¹)	MAP (mmHg)	SPO ₂ (%)	ETCO ₂ (%)
Case 1	Beginning of the process	88	93	96	41
	5 th min	83	78	98	38
	10 th min	78	80	96	35
	20 th min	81	79	96	34
	30 th min	77	75	93	38
	End of the process	75	77	97	34
Case 2	Beginning of the process	91	105	98	45
	5 th min	86	87	96	41
	10 th min	78	84	97	44
	20 th min	81	78	99	40
	30 th min	79	77	98	41
	End of the process	78	78	98	44

PHR: peak heart rate; MAP: mean arterial pressure; SPO₂: pulse oximeter; ET-CO₂: end-tidal CO₂; min: minute

Figure 2. Jet ventilation catheter passed through a stenotic lumen

After receiving written informed consent from the patient, she was taken to the operating room, and she was monitored. Following the establishment of vascular access, balanced electrolyte solution was initiated. The patient weighed 65 kg, and induction was provided with 2 mg of midazolam, 125 µg

kg⁻¹ of fentanyl, 150 mg of propofol and 14 mg of mivacurium. Tracheoscopy was performed according to the plan and method defined in the first case. Here, granulation tissues were found around the tracheostomy cannula. Thereupon, the excision of granulation tissue with CO₂ laser microsurgery was planned for the patient. When the tracheal cannula was removed, a serious stenosis was detected due to the granulation tissue in the trachea. An infraglottic JV catheter was inserted from the stoma to the trachea and ventilation was started after doing JV settings. One minute before resection with the CO₂ laser, it was passed to the laser mode adjusted as FiO₂: 0.35. The pressure of CO₂ was intermittently evaluated with ETCO₂ monitoring. The maintenance of anaesthesia was provided with propofol at a rate of 6–10 mg kg⁻¹ h⁻¹ and remifentanyl at a rate of 0.05–0.25 µg kg⁻¹ min⁻¹. Additionally, 8 mg of dexamethasone, 50 mg ranitidine, 10 mg of metoclopramide, and 1 mg kg⁻¹ prednisolone were administered.

Without the development of any complication during the intraoperative period in the patient, SpO₂ was between 96% and 99%, and ETCO₂ was between 40 mmHg and 45 mmHg (Table 1). After completing the process, the initial settings were switched back in JV with FiO₂ of 100%. When spontaneous respiration started after ventilating for 1 min (frequency 300/min, DP: 0.8 bar), the JV catheter was removed. Because respiration was normal and SpO₂ ranged between 97% and 99% in the patient who completely regained her consciousness and cooperation, she was taken to the recovery room with an oxygen support mask without recannulating. When no respiratory problem was observed during the follow-ups in the recovery room, she was decannulated and transferred to the clinic.

Discussion

Subglottic stenosis occurring with the proliferative growth of diseased tissue towards the larynx and trachea in WG can cause airway obstruction and require intervention. This situation is more common in the 4th and 5th decades of life, and its symptoms range from cough and shortness of breath to severe stridor that is progressively life-threatening. Approximately 16–23% of these patients develop subglottic stenosis, and this generally progresses with fibrosis and oedema leading to stenosis that is 3–4 cm below the vocal cord. In a series comprising 7 patients, which was conducted by Alaani et al. (4), in the case of active disease, tracheostomy was recommended as the first surgical treatment choice in addition to medical treatment for resolving respiratory failure. In both our patients, surgical tracheostomy was performed due to the development of serious respiratory distress and stridor associated with the progression of the disease while medical treatment was being received.

The progression of the disease continues in patients with tracheostomy, and the formation of granulomas surrounding the tracheostomy cannula and extending downwards causes serious stenosis in the airway.

Accordingly, a new surgical treatment is needed within the process. Lebovics et al. (5) reported the presence of subglottic stenosis in 25 of 158 patients with WG, and they specified that the lesion was so serious that some interventions such as dilatation, laser resection or laryngotracheoplasty would be needed in 16 of these patients. In both our patients, subacute deterioration on the basis of chronic disease and serious inspiration difficulty despite tracheostomy developed, and a new evaluation was necessitated. Firstly, the exact locations and degrees of lesions were determined with the help of images obtained through CT, and tracheobronchoscopy was performed under general anaesthesia for the intervention plan of treatment.

During general anaesthesia applied in tracheobronchoscopy, which is performed for the evaluation of the lesion and for planning surgical intervention and then applied in the intervention for the airway, airway control is important. Gao et al. (6) observed tracheal stenosis and dyspnoea in 12 of 15 patients having a mass in the trachea. In these patients, standard orotracheal intubation was preferred for stenoses, causing narrowing less than 50% in the trachea lumen, and the tracheostomy was opened in the mass located in the upper region of the trachea. On the other hand, in patients with a mass located in the lower region of the trachea and with serious respiratory distress and hypercarbia, adequate oxygenation could not be provided with the conventional technique. For airway control in these patients, extracorporeal circulation was first applied, and passing an endotracheal tube through a stenotic lesion with a fibre-optic bronchoscope was then implemented as an alternative technique. As another method, Xu et al. (7) reported that an adequate surgical site and appropriate surgical environment were provided with no development of any complication by protecting spontaneous respiration under infusion with propofol and remifentanyl without endotracheal intubation in 31 patients who were to undergo upper airway surgery.

In the 10-year observational study, Jaquet et al. (8) brought the subglottic JV forward as an alternative to the choices of intubation with a small endotracheal tube, tubeless ventilation with the protection of spontaneous respiration, and apneic oxygenation in the pathologies causing stenosis in the larynx. The JV technique can be evaluated as an alternative for the resection of granulation tissue with a CO₂ laser owing to its thin catheter structure and feature of laser resistance (9). In our patients, the JV technique was used for the resection of granulation tissue, which surrounded the existent tracheostomy, with a CO₂ laser.

Conclusion

In the advanced stages of WG, the infraglottic JV technique is a good option among techniques that can be used for enabling airway and for performing gas exchange during surgery conducted for granulations in the upper airway. In this technique, the thin structure of the catheter offers appropri-

ate vision for surgery and good working conditions during resection with the CO₂ laser and provides safe airway management and gas exchange for anaesthetists.

Informed Consent: Written informed consent was obtained from patients who participated in this case.

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References

1. Yazıcı ZM, Kayhan FT, Erdur Ö, Erol BS. Granülomatoziste KBB tutulumu. KBB ve BBC Dergisi 2009; 17: 62-5.
2. Zycinska K, Wardyn K, Zielonka TM, Nitsch-Osuch A, Zarzycki S, Demkow U, et al. Subglottic and tracheal stenosis due to Wegener's granulomatosis. Adv Exp Med Biol 2013; 755: 221-4. [\[CrossRef\]](#)
3. Grillo HC, Donahue DM, Mathisen DC, Wain JC, Wright CD. Postintubation tracheal stenosis. J Thorac Cardiovasc Surg 1995; 109: 486-92. [\[CrossRef\]](#)
4. Alaani A, Hogg RP, Drake Lee AB. Wegener's granulomatosis and subglottic stenosis: management of the airway. J Laryngol Otol 2004; 118: 786-90. [\[CrossRef\]](#)
5. Lebovics RS, Hoffman GS, Leavitt RY. The management of subglottic stenosis in patients with Wegener's granulomatosis. Laryngoscope 1992; 102: 1341-5. [\[CrossRef\]](#)
6. Gao H, Yi J, Huang YG. Airway management and anesthesia for tracheal masses in 15 patients. Zhongguo Yi Xue Ke Xue Yuan Xue Bao 2013; 35: 322-6.
7. Xu J, Yao Z, Li S, Chen L. A non-tracheal intubation (tubeless) anesthetic technique with spontaneous respiration for upper airway surgery. Clin Invest Med 2013; 36: 151-7.
8. Jaquet Y. Complications of different ventilation strategies in endoscopic laryngeal surgery. Anesthesiology 2006; 104: 52-9. [\[CrossRef\]](#)
9. Biro P. Jet ventilation for surgical interventions in the upper airway. Anesthesiol Clin 2010; 28: 397-409. [\[CrossRef\]](#)