## CASE REPORT

# Repetitive postoperative extubation failure due to dynamic inspiratory airway collapse concomitant with subglottic stenosis in a patient who previously underwent tracheostomy.

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**Abstract : Background : This report describes a case of dynamic inspiratory airway collapse concomitant with subglottic stenosis in a patient who previously underwent tracheostomy that led to repeated post-operative extubation failure. Case presentation : A 43-year-old woman who had undergone tracheostomy 25 years previously was admitted to our intensive-care unit (ICU) after coronary artery bypass graft surgery. On postoperative day (POD) 0, she was extubated, but stridor was observed. We suspected upper airway obstruction and she was therefore reintubated. Before reintubation, urgent laryngotracheoscopy revealed dynamic inspiratory airway collapse and obstruction concomitant with subglottic stenosis. Preoperative computed tomography showed mild subglottic stenosis. Although intravenous corticosteroids were administered to prevent tracheal mucosal edema and a cuff leak test was confirmed to be negative, she developed extubation failure on POD6. On POD12, we performed tracheostomy to reduce mechanical irritation from the endotracheal tube. Mechanical ventilation was withdrawn and she discharged from the ICU. On POD33, her tracheostomy tube was removed and she remained clinically asymptomatic. Conclusions : We should be aware of the history of tracheostomy, especially at high tracheostomy sites, even in the absence of respiratory symptoms as risk factors for dynamic inspiratory airway collapse concomitant with subglottic stenosis contributing to repeated respiratory failure after extubation. J. Med. Invest. 70 : 301-305, February, 2023**

***Keywords*** **:** *subglottic stenosis, tracheostomy, postoperative extubation failure*

## INTRODUCTION

Tracheal stenosis and tracheomalacia are the most common late complications after tracheostomy, and can be life-threatening (1, 2). Some degree of tracheal stenosis almost always occurs after tracheostomy, and 3–12% of patients require intervention for stenosis (1-3). Post-tracheostomy tracheal stenosis is quite difficult to diagnose because only 1–2% of patients develop clinically detectable symptoms (3-5). The internal diameter of the trachea is often considerably reduced before any symptoms become apparent and clinical symptoms are not sufficient to assess the airway patency, which may present weeks to years after tracheostomy (2). Severe tracheal stenosis may lead to unexpected airway management difficulties, including difficult intubation and ventilation at the induction of general anesthesia (5, 6). Furthermore, it may be a risk factor for post-extubation respiratory failure due to the further aggravation of airway narrowing and tracheal wall weakness caused by tracheal edema after extubation (5-8).

We herein report a rare case of repeated post-extubation respiratory failure in a patient who had undergone tracheostomy without any respiratory symptoms 25 years previously, mainly due to dynamic inspiratory airway collapse concomitant with subglottic stenosis, which was exacerbated by mechanical irritation from the endotracheal tube (ETT) after cardiothoracic surgery.

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## CASE PRESENTATION

A 43-year-old woman with a two-week history of fever, cough and dyspnea on exertion was admitted to our intensive care unit (ICU). Her relevant medical history included tracheostomy for herpes simplex encephalitis 25 years previously, and she had undergone myomectomy under general anesthesia 7 years previously. According to her medical records, her trachea was intubated with an ETT (inner diameter [ID] 7.0 mm) for general anesthesia and there were no respiratory problems in the perioperative period. Her family history showed that her mother had been diagnosed with systemic lupus erythematosus.

On preoperative computed tomography (CT) of the trachea, the subglottic tracheal diameter at the most stenotic site (from the cricoid cartilage to the first tracheal ring), measured using electronic calipers, had a depth of 14.3 mm and a width of 9.5 mm, which was 66% of the subglottic tracheal dimensions at other sites (Figure 1). The severity of subglottic stenosis is assessed by the Myer and Cotton grading system, and the patient had mild tracheal stenosis (Grade 1). Blood tests showed elevated inflammatory markers and cardiac enzymes (white blood

cell count $4.5 \times 10^3/\mu L$, C-reactive protein 8.9 mg/dL, creatine kinase 501 IU/L, creatine phosphokinase MB isoenzyme 47.3 IU/L). Electrocardiography showed ST depression on Ⅰ, Ⅱ, aVF and V2-6, and ST elevation on aVR and V1-2. Echocardiography showed diffuse hypokinesis, and coronary angiography showed 90% stenosis of the left main coronary trunk. Contrast enhanced CT of the thorax, abdomen and pelvis revealed thickening of the arterial wall from the ascending aorta to the aortic arch. Thus, the patient was diagnosed with ischemic cardiomyopathy due to aortitis syndrome.

Emergent off-pump coronary artery bypass grafting surgery (OPCAB) was performed under general anesthesia. At the induction of general anesthesia, there were no episodes of difficult intubation or resistance during the insertion of the ETT (ID 7.0 mm outer diameter [OD] 9.6 mm ; PORTEX®, Smith Medical Inc., Minneapolis, MN, USA). After successful intubation, the cuff was inflated to 25 $cmH_2O$. The procedure was completed without any adverse events with a total operation time of 4 hours, 10 minutes and an anesthesia time of 5 hours, 30 minutes. After surgery, she was admitted to the ICU on mechanical ventilation.

On postoperative day (POD) 0, the patient was extubated after an evaluation of respiratory and hemodynamic status and confirmation that her condition met the criteria for extubation. Soon after extubation, she was fully alert, but she appeared short of breath and inspiratory stridor was auscultated, which led to hypoxia. She was therefore reintubated with an ETT (ID 7.0 mm, OD 9.6 mm). After reintubation, her oxygenation immediately improved. We therefore considered that her post-extubation respiratory failure may have been caused by laryngeal edema. On the following day (POD1), we attempted re-extubation after loading methylprednisolone (20 mg every 4 hours before extubation, total 60 mg) and confirmation that a cuff leak test was negative ; however, the same symptoms appeared. We suspected anatomical abnormality of the upper airway. Urgent laryngotracheoscopy revealed subglottic stenosis. It appeared as an A-shaped deformity with residual suture material, resulting in malacia with dynamic inspiratory airway collapse and obstruction. There was no obvious laryngeal edema ; however, vocal cord dysfunction was observed. We speculated that this dynamic airway collapse concomitant with airway narrowing may have been due to the formation of granulation soft tissue induced by residual suture material, tracheal mucosal edema induced by mechanical irritation from the ETT, and weakening of the tracheal wall by previous tracheostomy (Figure 2). Her trachea was subsequently reintubated using a smaller ETT (ID 6.5 mm and OD 8.9 mm). On POD6, the patient was extubated again after achieving a 4.5 kg reduction in body weight with the administration of diuretics to reduce volume overload, loading methylprednisolone at the same dose as in the previous extubation, with the

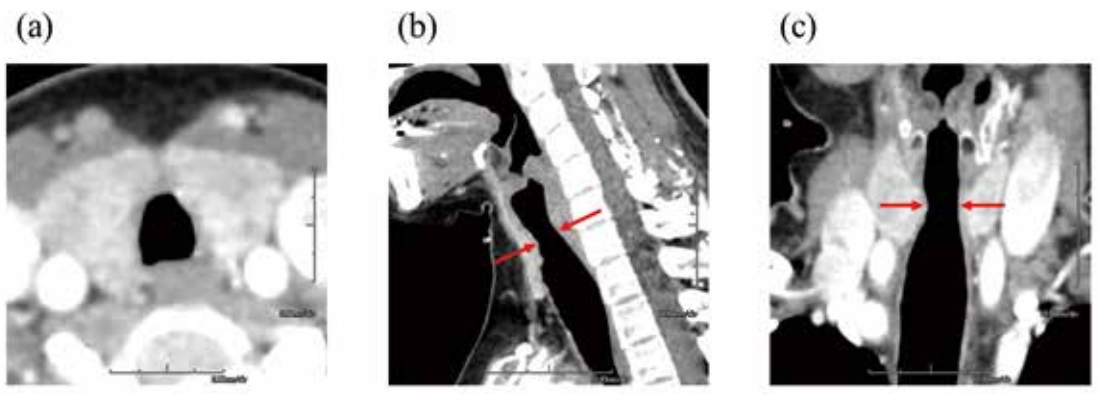


Figure 1. Preoperative computed tomography of the trachea showing subglottic stenosis
a) Axial view. b) Sagittal view. c) Coronal view
The red arrows indicate the most stenotic site of the trachea.

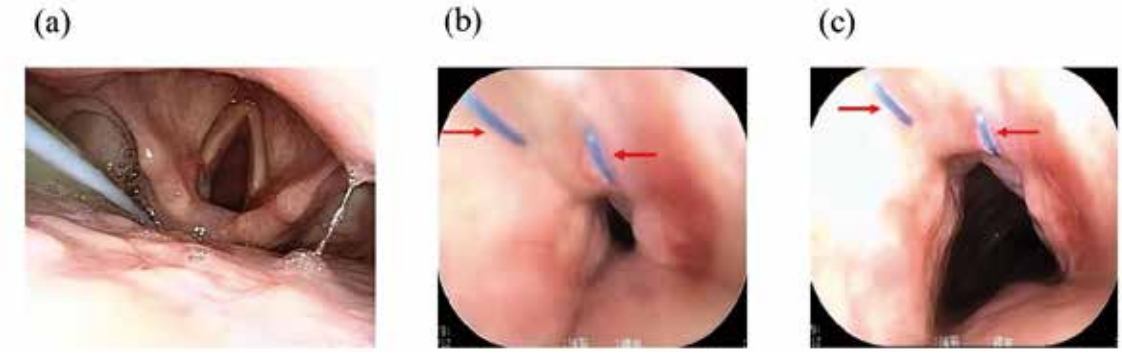


Figure 2. Laryngotracheoscopy showing subglottic stenosis and dynamic inspiratory airway collapse after extubation on POD 1.
a) : Vocal cord without any swelling or edema. b) The inspiratory phase. c) The expiratory phase

There was subglottic stenosis without laryngeal edema, which led to dynamic inspiratory airway collapse and obstruction. Residual sutures thread with granulation tissue was present at the upper side of the tracheal wall (b, c : red arrow).

patient in the head-up position, and confirmation that a cuff-leak test was negative. However, at 12 hours after extubation, she required reintubation again due to dyspnea and difficulty clearing secretions. On POD12, we performed surgical tracheostomy below the subglottic stenotic lesion to avoid excessive mechanical irritation from the ETT and the residual suture material was completely removed. Mechanical ventilation was withdrawn promptly after tracheostomy and the patient was discharged from the ICU by the end of the following day.

After discharge from the ICU, she was treated with inhaled betamethasone and an otorhinolaryngologist performed laryngotracheoscopy to evaluate the site of subglottic stenosis. Her subglottic stenosis had gradually improved (Figure 3), and on POD33 her tracheostomy tube was successfully removed without any symptoms associated with tracheal stenosis.

## DISCUSSION

We reported a rare case of repeated post-extubation respiratory failure in a patient with a history of tracheostomy who had not experienced respiratory symptoms for 25 years. It mainly occurred due to subglottic stenosis, which was exacerbated by mechanical irritation from the ETT after OPCAB. The pathophysiology of her respiratory failure had a unique characteristic showing a dynamic inspiratory airway collapse concomitant with subglottic stenosis at the site of the stoma in the previous tracheostomy. This is the first case report to describe that a high tracheostomy site may be a risk factor for post-stomal tracheal stenosis leading to dynamic inspiratory airway collapse and repeated post-extubation respiratory failure.

Tracheal stenosis is considered to be an iatrogenic airway injury after tracheostomy or prolonged intubation and remains a serious problem in critically ill patients (1, 2, 9). However, mild tracheal stenosis may produce no symptoms (e.g., cough, dyspnea and stridor). Exertional dyspnea occurs when the internal diameter of the trachea is reduced to $\leq 10$ mm, and dyspnea at rest or stridor occurs when it is reduced to $\leq 5$ mm (1). If the internal diameter of the trachea is reduced by 50–75% or narrowed to $\leq 5$ mm, symptoms occur and we can finally suspect the presence of tracheal stenosis (1, 2, 5, 10). Previous studies reported that clinical manifestations of tracheal stenosis often developed within 2 months after tracheostomy or intubation (1, 2), however, two studies reported that a patient developed upper airway stenosis after being asymptomatic for 18–20 years (3, 11). In this case, the patient had a history of tracheostomy and had been asymptomatic for 25 years after tracheostomy. Her tracheal diameter at the most stenotic site was estimated to be 14.3 mm deep and 9.5 mm wide. These dimensions would be considered to be non-symptomatic (1). Therefore, when investigating the presence of tracheal stenosis after tracheostomy or the risk for post-extubation respiratory failure, clinical manifestations are insufficient. We should be aware of the history of tracheostomy as a risk factor for post-extubation respiratory failure, even if a patient has not experienced any respiratory symptoms for a long time after tracheostomy.

Post-tracheostomy tracheal stenosis commonly occurs at the levels of the subglottic or stomal site, which is caused by inadequate incision and damage of the 1st tracheal ring or cricoid cartilage during tracheostomy procedures (1, 2, 9, 12). Typically, there is abnormal wound healing with excessive granulation soft tissue formation around the stoma site and an associated cartilage fracture or tracheomalacia of the tracheal wall (1, 2, 9). In particular, a high tracheostomy site is associated with a higher risk of tracheal stenosis and malacia, because it may damage the cricoid cartilage, which is the only circular cartilage in the upper airway (9, 13). In tracheostomy procedures, the presence of residual suture material in granulation tissue, as seen in our patient, may potentially contribute to cicatricial subglottic stenosis (14).

Furthermore, dynamic airway collapse at the subglottic stenotic site was observed in this patient during the inspiratory phase. Tracheomalacia is commonly characterized as weakening of the tracheal wall leading to dynamic expiratory airway collapse because the malacic site is typically located in the intrathoracic cage (1, 2). However, when the malacic site is located in the extra-thoracic cage, as seen in our patient, weakening of the tracheal wall due to cartilage damage may lead to dynamic inspiratory airway collapse because it is not affected by the positive

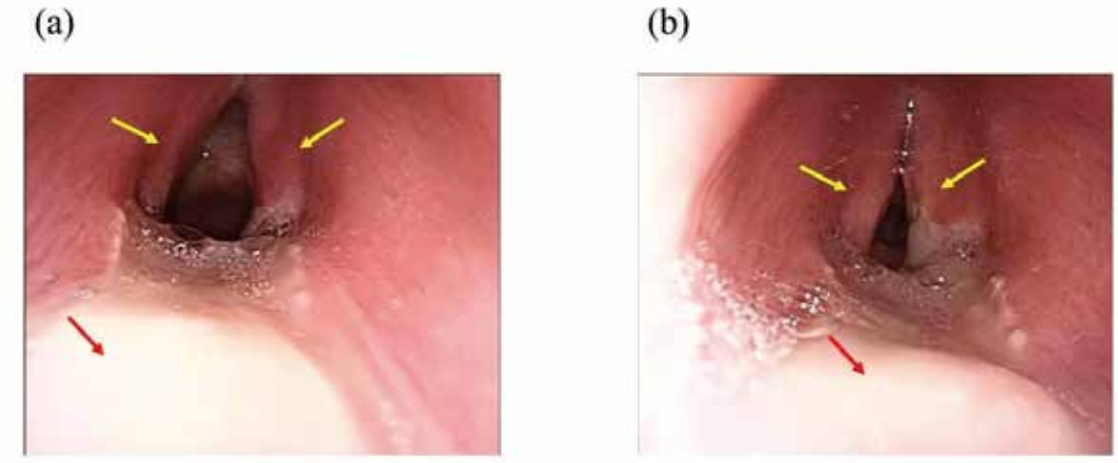


Figure 3. Subglottic site after tracheostomy.
a) The inspiratory phase. b) The expiratory phase.

Laryngotracheoscopic observation from the tracheal stoma and vocal cord (yellow arrow) after removal of the tracheostomy tube. The subglottic stenosis had improved and dynamic airway collapse was not seen during inspiration (red arrow). The patient's vocal cord dysfunction also improved (yellow arrow).

intrathoracic pressure during forced expiration, but rather is affected by forced inspiratory flow due to the Bernoulli phenomenon and is sucked into the central airway (2, 15). According to a previous study, the flow-volume curve of the pulmonary function test, which is sensitive in identifying the location and characteristics of airway narrowing, revealed that lesions with dynamically variable narrowing were subject to transmural pressure gradients, so that when the narrowing lesion was located in the extra-thoracic cage, it appeared as blunting of the inspiratory flow-volume loop and when the narrowing lesion was located in the intrathoracic cage, it appeared as blunting of the expiratory flow-volume loop (16).

Tracheal stenosis may also occur at the site of the tracheostomy tube cuff, caused by inadequate management of the cuff pressure (1, 2). Tracheal stenosis below the stoma typically results from ischemia of the tracheal mucosa due to the tracheal tube cuff pressure exceeding the perfusion pressure (1, 2). Because the stenotic site in our patient was located in the subglottic region, and not at the site of the tracheal cuff, we suspected that the subglottic stenosis and mucosal edema were exacerbated by mechanical irritation from the ETT.

Several predisposing factors for the development of tracheal stenosis have been described in the literature, including : high tracheostomy site, prolonged intubation, excessive corticosteroid use, advanced age, female sex, severe reflux disease, cardiovascular disease, obesity, autoimmune and systemic inflammatory diseases, and local radiation therapy (4, 9, 13). This patient's risk factors for subglottic stenosis included a high tracheostomy site, female sex, and comorbidities of cardiovascular disease and systemic inflammatory disease. Accordingly, this patient may have been more vulnerable to mechanical irritation from the ETT and the subsequent development of tracheal mucosal edema in comparison to a healthy subject.

There are several strategies for the prevention and management of extubation failure due to cicatricial subglottic stenosis and mucosal edema. Medical strategies for subglottic swelling include corticosteroids, diuretics, IV fluid restriction, head-up positioning, nebulized epinephrine, and occasionally reintubation with a smaller ETT (17). Surgical strategies for cases in which medical management is ineffective include laser resection, rigid bronchoscopic dilation, removal of granulation tissue with a biopsy forceps and stent implantation (1, 2, 4, 18). In this case, conventional medical management, including corticosteroid, diuretics and intubation using a smaller ETT, was not effective. Prolonged ETT placement was considered disadvantageous because of the potential for development of mucosal edema and further narrowing of the trachea after tracheal extubation. Because the preoperative tracheal stenosis was mild, and there were no symptoms of respiratory failure for 25 years, we considered that removing the ETT, which was the source of mechanical irritation of the tracheal mucosa, would be effective for improving the patient's subglottic stenosis and may have led to successful extubation. Performing surgical tracheostomy to avoid excessive mechanical irritation and bypass the stenotic site enabled mechanical ventilation to be withdrawn earlier and for the airway patency to be maintained.

## CONCLUSION

The patient's history of tracheostomy with asymptomatic airway stenosis that had been present for more than 20 years may have been a risk factor for repeated post-extubation respiratory failure and a post-stomal site after tracheostomy may cause malacia that is characterized by a dynamic inspiratory airway collapse and obstruction. In particular, the patient may be at higher risk for this phenomenon when the stomal site from the previous tracheostomy is higher than that of the usual approach. Preoperative assessment of airway patency and risk factors for extubation failure and preparation for unexpected airway emergencies at the time of extubation are mandatory for patients with a history of tracheostomy. Tracheostomy may be potentially effective for post-tracheostomy tracheal stenosis as it avoids mechanical irritation from the ETT and may lead to successful extubation.

## CONFLICT OF INTERESTS

All authors have no conflict of interests

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