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Membranous tracheal stenosis in a patient with anorexia nervosa and self-induced vomiting- challenges in securing the airway

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ABSTRACT

We report a rare case of acquired membranous tracheal stenosis in a patient with anorexia nervosa and a history of self-induced vomiting, but without a history of tracheal intubation or tracheostomy. A 50-year-old woman presented with difficulty in breathing and swallowing, self-expectoration, and impaired consciousness due to acute benzodiazepine intoxication. Bronchoscopic examination was performed after tracheotomy and placement of a tracheostomy tube failed to secure her respiratory tract and ventilation continued to deteriorate. A flap-like membranous structure was identified on the posterior tracheal wall, obstructing the tracheostomy tube. Physical compression of the membranous structure improved ventilation. Bronchoscopic examination is generally recommended prior to performing tracheostomy in patients suspected to have post-intubation tracheal obstruction. Based on our findings, we suggest that these examinations should also be performed in patients with conditions associated with chronic irritation of the respiratory tract, including those with a prolonged history of self-induced vomiting.

1. Introduction

Acquired tracheal stenosis has been reported as a complication affecting 10–22% of adult patients with prolonged tracheal intubation and tracheostomy [1,2]. Only 1–2% of these patients (an estimated 5 cases per million per year in the general population) have severe stenosis with symptoms of dyspnea and dysphagia [3,4]. We recently encountered a case of acquired tracheal stenosis in a patient with anorexia nervosa and a history of self-induced vomiting. However, this case is exceptional in that the patient had no history of tracheal intubation or tracheostomy.

2. Case report

Informed consent was obtained from the patient prior to the disclosure of any personal information and case details. A 50-year old woman (height: 155 cm, weight: 38 kg) was admitted to our hospital with difficulty breathing and swallowing, and impaired consciousness from suspected acute drug intoxication. The patient had a history of anorexia nervosa, for which she was receiving outpatient care from the psychiatric department and was treated with benzodiazepine therapy for 2 years. At admission, her mental status was E1V1M5 on the Glasgow Coma Scale (GCS; E – Eye opening, V – Verbal response, M – Motor response). Her vital signs were as follows: respiratory rate 24/minute, pulse rate 130/minute, systolic blood pressure 90 mmHg, and body temperature 38.3 °C. A benzodiazepine class drug was detected in the urine during triage. Suspecting acute benzodiazepine intoxication due to overdose, flumazenil was administered (0.5 mg intravenous bolus). Following flumazenil administration, her GCS mental status improved to E4V4M6. Aspiration pneumonia was also suspected, based on the findings of chest radiography, and symptoms of tachypnea, leukocytosis (7100 leucocytes/mL), neutrophilia, and elevated C-reactive protein (CRP: 11.5 mg/mL). The patient was admitted to our intensive care unit (ICU) for further monitoring and treatment for pneumonia by using intravenous tazobactam/piperacillin (4.5 g, three times a day for 14 days).

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While under continued observation, the patient showed no specific signs of anorexia nervosa (no detectable distortion of her sense of body image, no cognitive distortions such as the desire to lose weight, no recognizable signs of learned undesirable behavior in her eating pattern, no admitted or observed self-induced vomiting). However, she had poor food intake, which appeared to be related to difficulty in swallowing. Furthermore, she spoke softly and in short sentences as if in pain, and had frequent choking and coughing episodes resulting from fluid accumulation in her mouth and self-expectoration of phlegm (likely resulting from pneumonia). We also observed episodes of phlegm build-up at night, causing her blood oxygenation level (PaO2) to decrease to approximately 70%. Based on these observations, we suspected a neurodegenerative disorder such as amyotrophic lateral sclerosis, and referred the patient to the Department of Rehabilitation and Neurology for evaluation of swallowing function. However, no possible neurological causes for these symptoms were identified during the swallowing evaluation.

During recovery from pneumonia, the patient continued to have difficulty breathing and swallowing, as a result of frequent coughing and self-expectoration of phlegm. On the eighth day of hospitalization, in order to secure her airway while preserving vocal functions, we performed a tracheotomy under general anesthesia (intramuscular midazolam 10 mg, rocuronium 25 mg, fentanyl 0.1 mg) and inserted a tracheostomy tube (Portex® 8.0, Smiths Medical, UK). During the procedure, there was a sudden cease in ventilation. An immediate orotracheal intubation was performed and ventilation was normalized. Suspecting a mismatch of the tracheostomy tube with tracheal shape, we substituted the Portex® 8.0 tube for a flexible tracheostomy tube with an adjustable flange. However, using computerized tomography (CT) imaging, the placement of the tube was found to be too deep, and the tube location was adjusted upwards by 1 cm. As the tube position was adjusted, we again encountered a blockage in ventilation, and performed another orotracheal intubation to normalize ventilation. Computerized tomography imaging was performed again to check the tube positioning, and a tracheal membrane was observed adjacent to the tracheostomy tube (Fig. 1). A bronchoscopic examination was performed to confirm that this membrane was the cause of ventilation obstruction.

2.1. Bronchoscopy findings

We identified a membranous structure on the posterior tracheal wall, 2 cm cranial to the tracheal bifurcation. Additionally, the left main bronchus showed luminal collapse consistent with signs of tracheomalacia. The membranous structure appeared mobile, and acted like a flap to completely obstruct the tip of the tracheal intubation tube as it was pulled upwards. While observing the trachea with a bronchoscope, we applied pressure to the membranous substance on the posterior tracheal wall using the flexible tracheostomy tube, and waited several minutes for ventilation to normalize before concluding the operation.

On the 17th day of hospitalization, the patient was evaluated by an otolaryngologist and it was confirmed that the membranous structure in the tracheal wall was somewhat mobile, but not flexible enough to obstruct the tracheostomy tube. The patient was discharged and continued to receive outpatient treatment from the psychiatric department for anorexia nervosa.

3. Discussion

It is speculated that prolonged tracheal intubation exerts excessive or repeated mechanical pressure on the tracheal wall and irritates the tracheal mucosa [5–7], and that this irritation can result in membranous tracheal stenosis. When the capillary perfusion pressure increases to over 18–25 mmHg, mechanical edema and ischemia of the tracheal mucosa [4] could cause mucosal necrosis and erosions, which subsequently lead to the formation of a fibrous granuloma or scar in the tracheal mucosa. Indeed, a membranous flap-like morphology in tracheal obstructions has been described frequently in patients with a history of prolonged intubation or tracheostomy [5–12]. In the absence of a history of tracheal intubation, we suspect that self-induced vomiting was the cause of our patient's tracheal irritation and membranous tracheal stenosis. Patients with anorexia nervosa commonly insert their finger into their back of their throat to stimulate the pharynx, or apply pressure to their abdomen to stimulate the stomach, and induce vomiting. Furthermore, many patients generally conceal this behavior [13]. In this case, the patient denied that she had been inducing vomiting. The patient would typically avoid eye-contact when questioned about self-induced vomiting, and admitted to feeling nausea during meals. In addition, according to information obtained from her previous doctor, she had frequently self-induced vomiting for over a year. Based on the patient's statements and this information, we speculated that it was the external force from chronically pressurizing the respiratory tract that resulted in the formation of a membranous obstruction in the trachea. In addition, irritation by the vomitus and aspiration of gastric contents could also contribute to tracheal irritation and stenosis [6,8,14].

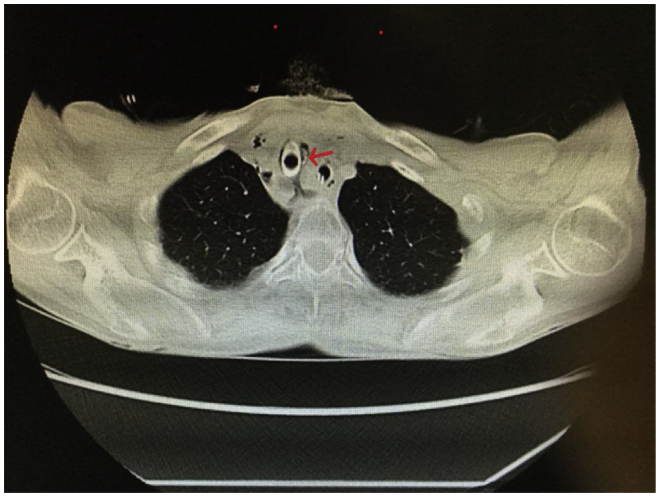

Fig. 1. Computerized tomography (CT) scan performed after tracheotomy showing the tracheal membrane (indicated by an arrow) obstructing the tracheostomy tube.

It is recommended that patients with suspected tracheal obstruction are initially evaluated using tracheal intubation and bronchoscopy, to assess the degree of damage to cartilage and determine an appropriate treatment algorithm [15]. Based on tracheotomy findings, various treatment options are recommended, such as reconstructive surgery of the trachea through a thoracotomy, laser ablation, stent placement, and dilation through the use of bronchoscopes or balloons [4]. We were not aware of any reports of membranous obstructions in the trachea of patients without a history of tracheal intubation; therefore, we did not perform an initial evaluation by tracheal intubation and bronchoscopy, but proceeded directly to tracheotomy, to secure the airway while preserving speech. Based on the challenges in diagnosing and managing the current case, we recommend that a bronchoscopy is performed to examine the respiratory tract in all patients where tracheal damage is suspected based on patients' medical history. In particular, respiratory or swallowing difficulties in patients with a

prolonged history of self-induced vomiting should be evaluated by bronchoscopy before performing tracheotomy.

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Conflict of interest

None.

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