Original Article

Management of esophageal stenting-associated esophagotracheal fistula, tracheal stenosis and tracheal rupture: a case report and review of the literature

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Abstract: Objective: Although the placement of esophageal self-expandable stents (SES) can effectively relieve dysphagia after radiotherapy in patients with esophageal cancer (EC), it may induce severe esophageal complications. This article reports a case of emergency endotracheal intubation in an EC patient who suddenly developed severe dyspnea two months after SES placement. Methods: Electronic bronchoscopy of the patient’s airway confirmed the diagnosis of esophagotracheal fistula, tracheal stenosis and tracheal rupture. Endotracheal intubation was successfully performed under the guidance of electronic bronchoscopy. Results: Dyspnea due to tracheal stenosis was relieved effectively by inserting the tracheal catheter to a proper place under the guidance of electronic bronchoscopy. Conclusion: Bronchoscopic examination is strongly recommended in EC patients who are highly suspected as having airway stenosis associated with esophageal stenting, for which endotracheal intubation under the guidance of bronchoscopy is suggested.

Keywords: Esophageal self-expanding stent, esophagotracheal fistula, tracheal stenosis, tracheal rupture, airway management

Introduction

Placement of esophageal self-expandable stents (SES) is a recommended option for the treatment of esophageal stenosis in the late stage of esophageal cancer (EC) because it can effectively relieve dysphagia due to esophageal stenosis [1]. Stent-associated esophageal respiratory fistula (SERF) is a serious post-SES complication that affects the airway safety and threatens the life of the patients. Intra-tracheal or intra-esophageal placement of SES to isolate the air passage for the sake of preventing pulmonary complications is the common treatment for SERF [2]. However, mal-introduction of the tracheal catheter into the esophagus and improper placement of the tracheal catheter are likely to occur during emergency tracheal catheterization. This article reports a case of successful tracheal catheterization in an EC patient who was confirmed as having SERF, tracheal stenosis and tracheal rupture in the context of review of the literature.

Case report

A 51-year-old male patient with a 2-year history of EC had received chemotherapy for a cycle and radiotherapy for two cycles. Four months later, he received SES placement in another hospital. After that, he complained of hoarseness of voice, chest stuffiness and suffocation that exacerbated after exertion, which affected his daily activities or awoke him at night. The symptom could be relieved by orthopnea. He coughed up yellow sputum with blood streaks. On 2014-02-05, he received anti-infection therapy for pulmonary infection in a local hospital. But as the treatment was not effective, the patient was transferred to our hospital for continuous anti-infection therapy. On March 5, the patient suddenly developed severe dyspnea,
which could not be relieved by orthopnea. After emergency consultation with the Department of Anesthesiology, emergency endotracheal intubation was performed. Physical examination showed the following: orthopnea, mask oxygen inhalation, no subcutaneous emphysema, HR 126 bpm, SaO₂ 87%, and communication with gestures. Based on the review of the medical records and 02-14 CT images in another hospital (Figure 1) in the context of our clinical findings, tracheal rupture and stenosis were highly suspected. After discussion with the family and the responsible physician, informed consent about endotracheal intubation was signed. The electronic bronchoscope, video laryngoscope and other emergency instruments and materials were prepared. With the patient assuming a sitting position, midazolam (1 mg) and the mixture of lidocaine and ephedrine were administered intravenously (IV) for nasal mucosa convergence and anesthesia. The bronchoscope was inserted through the left nostril. After ensuring that there was no abnormality in the supraglottic structure, the patient was instructed to take a deep breath; the bronchoscope was inserted the moment the glottis was open. Bronchoscopy (Figure 2) showed the air passage was obviously abnormal, with the SES protruding into the trachea (C); the opening of the esophagus being in the trachea (D); and the opening of the trachea being compressed flat by the SES (E), where air flow and secretion were seen flowing out of the flattened trachea. The bronchoscope was inserted accurately through the opening of the trachea (E), finding that the tracheal membrane was rupture, where the SES was seen clearly. The ruptured part extended about 2 cm above the carina (Figure 3). Through the bronchoscope, a 6.5 reinforced tracheal catheter was inserted, with the opening of the catheter located at about 1 cm above the carina (Figure 3) reaching a depth of 28 cm in the nostril. The bronchoscope was then withdrawn, and the cuff of the tracheal trocar was inflated. Oxygen was inhaled through the tracheal catheter. Two minutes later, the patient’s SaO₂ rose to 96%, HR dropped to 90 bpm, and the patient felt much comfortable. With the patient lying flat in a supine position, a gastric tube was inserted through the right nostril under video laryngoscopy. After seeing gastric fluid flowing out, the tube was fixed. Five days later, a main tracheal and bronchial stent (Y type) was placed under local anesthesia, and the patient was discharged spontaneously.

Discussion

SES placement is a recommended option for the treatment of esophageal stenosis in the late stage of EC, as well as in patients with benign esophageal stenosis and EERF. SES placement is frequently used because of the obvious outcome. Bick et al [3] showed that SERF occurred in 4% (16/397) of their patients 5 months after SES placement. SERF occurred in the proximal segment of the esophagus in
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6% cases, in the middle segment in 14% cases, and in the distal segment in no case. The incidence of SERF is similar in patients with benign esophageal diseases. There is no evidence of metastasis occurring at the SERF site, suggesting that the effect of SES compression on the esophageal soft tissue and the action of tracheal pressure on the occurrence of SERF is greater than that of malignancy or benignity of the disease itself. Bethge et al [4] reported the microscopic evidence of cellular necrosis and fibrosis in the SES-impacted tissue, suggesting that the occurrence of SERF is associated with tissue ischemia and necrosis. SERF in patients with malignant esophageal tumors can be managed with esophageal or tracheal stent placement. However, it may increase the local pressure on the tissue, causing an even greater fistula opening [3]. Malignancy of the disease itself is an important factor contributing to the occurrence of SERF [5].

Patients with SERF often present with non-specific respiratory and esophageal complications, and the duration from the symptom onset to the confirmed diagnosis is about 3 weeks [3]. In some cases SERF cannot be confirmed until the diagnosis of pneumonia, and therefore mis- and mal-diagnosis are likely to occur. In our patient, dyspnea and orthopnea were present after surgery, and therefore we suspected him as having severe airway stenosis, which was later confirmed by bronchoscopy. The duration from admission in the local hospital after the onset of symptoms to the referral to our hospital where the diagnosis was confirmed was 4 weeks, or 3 days after admission into our hospital.

Radiotherapy is a safe and effective treatment for inoperable patients with EC [6]. However, the history of radiotherapy is the most important risk factor contributing to the occurrence of SERF. The risk of SERF in patients with a history of radiotherapy is 9.4 fold that in patients without receiving radiotherapy [3], which is similar to the result of Park et al [7]. Radiotherapy itself can induce ERF [8]. Radiotherapy and subsequent stent placement seem to play a significant synergistic effect on the occurrence of SERF. Studies [9] have demonstrated that radiotherapy can reduce the tissue elasticity and perfusion between the esophagus and trachea, and increase the tendency of necrosis due to increased pressure on the tissue. Cardiovascular diseases and diabetes themselves can lower tissue perfusion and unfavorably affect wound healing [3]. As the occurrence of SERF is relatively high and the prognosis is poor, other therapeutic options can be considered in patients with EC occurring in the proximal or middle segment of the esophagus. Park et al [7] observed 208 EC patients who received radiotherapy after covered-stent placement for malignant esophageal stenosis and found airway-related complications in 23 (11%) patients, including ERF in 18 (78.3%) cases, airway stenosis in 3 (13.0%) cases, and both in 2 (8.7%) cases. There is a significant difference in the occurrence of airway complications between radiotherapy patients and non-radiotherapy ones, but there is no significant difference between patients receiving radiotherapy before and after stent placement. The interval of stent-related complication occurrence and the duration of survival of patients who received radiotherapy before stent placement are significantly shorter than those in patients receiving radiotherapy after stent placement. Tracheal rupture usually occurs in patients with head and neck traumatic injuries [10], and is also the complication related to endotracheal intubation [11], tracheoscopy and tracheostomy. Endotracheal intubation is the common iatrogenic factor contributing to tracheal rupture [12]. Miñambres et al [10] observed tracheal rupture in 182 postoperative patients and found that emergency endotracheal intubation was the main reason for tracheal rupture, the incidence being 3 fold as high as that with selective endotracheal intubation. In addition, it is also the only factor related to patient death in this setting. Tracheal rupture is a rare and severe complication after...
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endotracheal intubation, the clinical diagnosis of which is relatively difficult. The mortality of tracheal rupture is as high as about 22% [13]. The most common clinical symptom is dyspnea and shortness of breath [14]. Percutaneous emphysema is the most obvious sign [10]. In our patient, no endotracheal intubation was performed and no obvious subcutaneous emphysema was observed, which may be the objective reason for the previous mis-diagnosis of tracheal rupture. Bronchoscopy is the gold standard for the diagnosis of tracheal rupture because it is able to display the location, nature and size of rupture. In recent years, chest CT scan has been more widely used in the diagnosis of suspected tracheal rupture due to convenience and high sensitivity [15]. Dyspnea in our patient was mainly due to protrusion of the subglottal stent into the air passage, causing compression on the trachea, severe subglottal stenosis and ERF. The reason for the absence of subcutaneous emphysema in our patient may be because the site of tracheal rupture was blocked by the stent so that air could not flow freely to other tissues. Tracheal rupture in this patient may be related to radiotherapy and/or ischemic necrosis of the tracheal membrane tissue due to the pressure of the esophageal stent. The main goal of treatment for tracheal rupture is to maintain the integrity of the airway and avoid complications. Tracheal rupture can be treated surgically or non-surgically [16]. In patients whose clinical status is relatively stable without severe dyspnea, conservative therapy may provide a relatively good prognostic outcome [17], which includes mechanical ventilation support, oxygenation, maintenance of fluid and electrolyte balance, prevention of infection, sedation and analgesia, and cardiothoracic nursing [16]. Bronchoscopic examination is recommended for patients at risk of tracheal rupture whose clinical status is unstable [18]. Endotracheal intubation should be performed under the guidance of bronchoscopy. Construction of an artificial air passage by avoiding the rupture trachea may prevent air leakage of the air passage and relieve the symptom of dyspnea. In the present case, we performed endotracheal intubation after confirmation of the diagnosis by bronchoscopy. As a result, the patient’s symptoms of hypoxia were improved significantly. We do not recommend using the conventional method of endotracheal intubation in such patients for the sake of avoiding exacerbating the existing tracheal injury or entry into the esophagus, thus endangering the safety of the patient. In summary, CT screening is suggested whenever possible in patients who present with unexplainable recurrences of pulmonary infection and dyspnea after esophageal stent placement before or after esophageal radiotherapy. Bronchoscopy is the gold standard for the diagnosis of ERF and tracheal rupture. In addition, endotracheal intubation under the guidance of bronchoscopy is the most safe and effective airway.

Disclosure of conflict of interest

None.

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