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Case Report

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Repeated Tracheostomy Tube Cuff Rupture: Tracheobronchopathia Osteochondroplastica

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Tracheobronchopathia osteochondroplastica (TPO) is a rare bening chronic disease that affects the lower part of the trachea and the upper part of the main bronchi and is characterized by multiple submucosal ossified and/or cartilaginous nodules projecting into the lumen of the airway. Here we report a case of TPO diagnosed during surgical tracheostomy by repeated tracheostomy tube-cuff rupture. Repeated cuff rupture during surgical tracheostomy occurred possibly because of bony and cartilaginous tissue in the tracheal wall. Many TPO patients are unable to be diagnosed because they are asympthomatic. In diagnosing TPO, the typical scene from Fiberoptic broncoscopy (FOB) is very pathognomonic and biopsy is not always necessary; FOB also helps to screen the tracheal area. If bed side percutaneous tracheostomy or surgical tracheostomy is planned, FOB may be routinely performed to diagnose TPO and/or to prevent complications from percutaneous or surgical tracheostomy.

Keywords: Tracheobronchopathia osteochondroplastica, surgical tracheostomy, repeated tracheostomy tube-cuff rupture

INTRODUCTION

Tracheobronchopathia osteochondroplastica (TPO) is a rare benign chronic disease involving the lower part of the trachea and the upper part of the main bronchi. It is characterized by multiple submucosal ossified and/or cartilaginous nodules projecting into the lumen of the airway (1-3). It was first reported by Rokitansky, Luschka, and Wilks in the 19th century (1-4). Its etiology, pathology, and natural history are still unknown (1-3). Two theories, Virchow's theory and metaplastic theory, have been postulated about the pathogenesis (1, 2, 5). TPO can be associated with chronic infections, inflammation, trauma, amyloidosis, silicosis, atrophic rhinitis, and allergic bronchopulmonary aspergillosis (1-3). The incidence of TPO ranges from 0.01% to 4.2% (2). Although nearly 400 cases have been reported worldwide, many TPO patients could not be diagnosed due to the unawareness of clinicians (2-4). The first presentation may be in the operating room with difficult intubation (1). Many patients affected by TPO are asymptomatic or manifest non-specific respiratory symptoms, such as chronic cough, dyspnea, hemoptysis, wheezing, and recurrent respiratory infections (2, 4, 5). It is usually diagnosed in the sixth or seventh decade of life, but it may also affect younger people (3-5). Here we present a case of repeated tracheostomy tube cuff rupture during surgical tracheostomy after we obtained consent from the patient's relatives.

CASE PRESENTATION

A 75-year-old-man, intubated by the 112 emergency medical service because of loss of consciousness after falling at home, was brought to the emergency department. From his medical history, it was found that he had been using warfarin because of the coronary artery bypass operation he had undergone. His physical examination showed that he was unconscious and had anisocoria (Right>Left). In his laboratory findings, the only abnormal result was a high international normalized ratio. His cranial computed tomography (CT) image showed acute subdural hematoma on the right side and shift on the midline. Following this, he was operated and then transferred to intensive care unit (ICU). On the 15th day of stay in the ICU, the patient was still unconsciousness. Therefore, tracheostomy was planned. However, because of a short neck, the Otorhinolaryngology Department planned to perform the operation. In the operating room, under general anesthesia, a horizontal incision was made through the second and third tracheal rings. From this incision (8.0-mm Portex Soft Seal cuff; Portex Ltd, Hythe, UK), a tracheostomy cannula was placed. Before placement of the cannula, the cuff was con-

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FIGURE I. Flatted tracheostomy cannula due to TPO

trolled and it was verified that it was working properly. However, after placement of the cannula, the cuff of the cannula was not working properly; it was flat. Hence, another tracheostomy cannula, the cuff of which was also controlled before placement, that was manufactured by the same company was placed, but it was also flat (Figure I). Subsequently, another tracheostomy cannula with a thicker cuff (8.0 mm Rüsch Ultra-Tracheoflex, Rüsch GmbH, Germany) was placed through the same incision. This cannula worked properly, and the cuff was not flat. During open surgical tracheostomy, which was performed by an ENT(Ear Nose Throat) doctor, the trachea was stony hard. Another issue was the repeated perforation of the tracheostomy tube cuff, which occurred even after changing the tube during the tracheostomy procedure. After the operation, the patient was transferred to the ICU, and he died I day later. No pathological specimen was sent for medical examination, and a cervical CT scan could not be obtained; these are the limitations of this case report.

DISCUSSION

The first case of TPO during surgical tracheostomy was reported by Nikandish et al. (I). Our case might be considered the second case of TPO during surgical tracheostomy because of the repeated perforation of the tracheostomy tube cuff during surgical tracheostomy. The repeated rupture of the tracheostomy cuff could be explained by the trauma to the cuff due to the presence of bony tissue in the tracheal wall. The presence of bony tissue can be explained by the calcification and ossification observed during surgical tracheostomy.

The manifestations of TPO may be either asymptomatic or non-specific respiratory symptoms (dry cough, productive cough, hemoptysis, and dyspnea, dryness of the throat, recurrent lower respiratory tract infection, and atelectasis) (2-5). TPO patients may also experience difficult intubation or repeated tracheal tube cuff rupture (I, 2). In cases where clinical signs are present, clinical symptoms depend on the location of occurrence of lesions (3). In our case, the patient's medical history was not clear; he had been using only warfarin for coronary artery disease, which may be the reason for him falling, along with respiratory reasons, such as pulmonary arrest, airway obstruction, or hypercapnic respiratory failure.

The X-ray image obtained during TPO has no characteristics (2). In the advanced phase of the disease, narrowing or displacement of the trachea is visible, with a marked irregular outline of its walls (3). No pathological appearance was reported in the X-ray of the present case. The results of pulmonary functional tests (PFTs) are within normal ranges in mild TPO patients. Results that are not within normal ranges in PFT indicate the severity of TPO (2, 3).

A CT scan may demonstrate the special finding of multiple submucosal calcified nodules involving the anterior and lateral walls of the tracheobronchus (2-5). The main specific images in fiberoptic bronchoscopy (FOB) are sessile submucosal calcified nodules protruding into the lumen of the trachea and main bronchi, maintaining the posterior wall intact (2). The pathognomonic feature that differentiates TPO from other disorders affecting the airways, such as amyloidosis, is the intact posterior wall of the trachea in TPO (3, 4). Although imaging studies may provide a clue to the diagnosis, bronchoscopy is the most definitive diagnostic test (5). To diagnose TPO, the typical scene in FOB is described as beaded, spiculate, rock garden, or cobble-stoned like nodules, which projected into the tracheobronchus lumen, sparing the posterior wall. very pathognomonic, and a biopsy is not essential since the typical appearance of FOB is enough to make a diagnosis. (2-4). The cartilage and ossification in the submucosa, calcification, and mucosal squamous metaplasia are the characteristic histopathological findings in bronchoscopic biopsy materials (2, 5). In our case, we had planned performing FOB and a cervical CT scan, but the patient died within 24 h (before they could be performed).

Medical treatment modalities are non-specific and are offered only for symptomatic patients (2). Several therapeutic modalities, including corticosteroids, airway humidity, laser ablation, surgical resection, mechanical debulking, stent implantation at the place of narrowing, cryotheraphy, radiotherapy, removal of nodules by forceps, and surgical treatment, are performed (2-5). The optimum treatment is controversial (4).

Our patient was intubated by the 112 emergency medical service and was brought to the emergency department. After an operation for his subdural hematoma, he was transferred to the ICU. He was monitored through a mechanical ventilator. On the 15th day of stay in the ICU, the patient did not gain consciousness; hence, tracheostomy was planned. Routinely, percutaneous tracheostomy is performed for such patients, but owing to the short neck of the patient, surgical tracheostomy was performed by an ENT doctor in the operating room under general anesthesia. If percutaneous tracheostomy had been performed, it could have been more complicated.

Many TPO patients cannot be diagnosed. To diagnose TPO, the typical scene in FOB is very pathognomonic, and a biopsy is not always necessary. FOB also helps to screen the tracheal area. If bedside percutaneous tracheostomy or surgical tracheostomy is planned, FOB may be performed routinely to diagnose TPO and/or prevent complications due to percutaneous or surgical tracheostomy.

Informed Consent: Written informed consent was obtained from patient's relatives.

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