中文題目: 氣管腺樣囊性癌-中央氣道阻塞的少見病因: 病例報告

英文題目: An Uncommon Etiology of Central Airway Obstruction – Tracheal adenoid Cystic

Carcinoma: A Case Report

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Introduction

Primary tracheal tumor is an uncommon etiology of central airway obstruction and may be life-threatening. Approximately 90% of primary tracheal tumors are malignant in adult patients. Adenoid cystic carcinoma (ACC) is the second common histopathologic type of primary tracheal malignancy, after squamous cell carcinoma (SCC). Here, we presented a case with tracheal ACC, which was ignored by chest radiography and initially was only treated with bronchodilator for wheezing. However, spirometry implied intra-thoracic central airway obstruction. The urgent contrast-enhanced chest computed tomography (CT) revealed tracheal tumor, consistent with the spirometry finding, and ACC was diagnosed by bronchoscopic tracheal tumor excision.

Case Presentation

A 66-year-old female never-smoker initially presented at our emergency department (ED) with exertional dyspnea, cough with little sputum, and wheezing for 1 week. She had history of endometrial cancer in her 54 years old, and abdominal total hysterectomy (ATH) and radiotherapy were done, without recurrence, but with post-operation related right lower extremity lymphedema. She also had obsessive-compulsive disorder (OCD) and suspected anxiety disorder. Initial physical examination revealed wheezing and right lower leg pitting edema. Venous blood gas revealed hypercapnia, and the other laboratory examination was unremarkable. No obvious abnormality of the initial chest x-ray was noted. After the bronchodilator inhalation therapy, her symptoms improved transiently and then she leaved from ED.

Two days later, dyspnea on exertion was still complained when she came back to our chest out-patient department. For further dyspnea and hypercapnia survey, spirometry was arranged. The bronchodilator test was negative. The flow-volume curve implied upper airway obstruction, variable intra-thoracic type. On the other hand, in the chest man opinion, trachea narrowing around the interclavicle area of the chest x-ray, taken 2 days before at ED, was considered. Then, the urgent contrast-enhanced chest computed tomography (CT) revealed a tracheal tumor with 3.7cm of greatest dimension. Multiple small lung nodules were also noted on high resolution computed

tomography (HRCT). Finally, adenoid cystic carcinoma of trachea was diagnosed by bronchoscopic tracheal tumor excision, and tracheal stent placement was also performed. Right lower lung video-assisted thoracoscopic surgery (VATS) wedge resection showed fibrotic thickening of alveolar walls and bridging fibrosis.

Discussion

Primary tracheal tumor is an uncommon etiology of central airway obstruction, as a potentially life-threatening condition, and approximately 90% of primary tracheal tumors are malignant in adult patients. The most common histopathologic type of primary tracheal malignancy is squamous cell carcinomas (SCCs), followed by adenoid cystic carcinomas (ACCs), and these 2 types composed about two-third of adult primary tracheal tumors.

The diagnosis of tracheal tumor may be delayed. It could be initially treated as asthma, chronic obstructive pulmonary disease, or bronchitis as its initial presentation of wheezing, dyspnea on exertion, or cough. Like our presented case, central airway obstruction, a potentially life-threatening condition, was ignored after symptoms improvement after bronchodilator inhalation therapy at ED. Tracheal tumor is difficult to diagnose according to chest radiography, and only a minority, about 18-28%, of tracheal tumors can be identified on chest x-ray.

Spirometry is a quick test and almost always prescribed for obstructive pulmonary disease survey when wheezing or dyspnea is complained. Central and upper airway obstruction can be identified according to flow-volume curves. A repeatable forced expiratory flow plateau, along with the lack of a forced inspiratory plateau, on the flow-volume curve implies a variable intra-thoracic central airway obstruction, like our presented case.

Chest CT with multiplanar reconstructions is the stander imaging study for tracheal tumor detection, staging, and pre-surgical planning. Before chest CT, besides clinical symptoms, spirometry is a good tool for upper airway obstruction evaluation, if the patient can cooperation with this examination.