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# A tracheobronchial adenoid cystic carcinoma incidentally detected by chest radiography and pulmonary function test

## Abstract

Tracheobronchial adenoid cystic carcinomas are rare tumors that progress slowly. Diagnosis by chest plain radiography is difficult, and suspecting the existence of a disease that causes stenosis in the airways is an important key for diagnosis. A 51-year-old woman referred to our hospital because of dyspnea on exertion. Chest plain radiograph showed a slight widening of the mediastinum. The flow–volume curve of respiratory function test revealed flat portions of the curve suggesting the central airway obstruction pattern. Chest CT confirmed the existence of tracheobronchial tumor, which was pathologically diagnosed as adenoid cystic carcinoma. Chest physicians and thoracic surgeons should be careful not to miss these trivial findings in the first inspections.

**Key words:** adenoid cystic carcinoma, tracheal tumor, pulmonary function test, flow-volume curve

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## Introduction

Adenoid cystic carcinoma has slow tumor growth, therefore, airway obstruction often progresses slowly [1–3]. Some patients initially have no symptoms or have non-specific mild symptoms such as cough and wheezing [2–4]. The tumor may develop along the tracheobronchial wall or into the central airspace without extending to surrounding structures. Therefore, diagnosis with plain chest radiograph is not easy even in advanced patients. Therefore, chest radiograph may not provide significant findings even in advanced patients. It has been reported that some subjects were misdiagnosed as bronchial asthma and consequently missed proper treatment [4, 5].

We present herein a case with a tracheobronchial adenoid cystic carcinoma incidentally detected by chest radiography and pulmonary function test. Although rare, trivial findings in these tests can lead to diagnosis, therefore, chest physicians and thoracic surgeons should be careful not to miss them.

## Case report

A 51-year-old female received chest radiograph in an annual workplace medical check-up. The patient had exertional dyspnea for a month prior to the check-up. She had no habit of smoking and no prior medical history of chest disease. She had the medical check-up for several years, but no abnormalities were pointed out in chest radiograph. In chest radiograph taken this time, the mediastinum structure widened to the right side compared to a chest radiograph taken last year for the medical check-up (Figure 1). The results of pulmonary function test were FVC 2.67 L (102.7% of the predicted value) and $FEV_1$ 0.94 L (41.8% of the predicted value). The flow-volume loop demonstrated flat portions of the curve suggesting the central airway obstruction pattern (Figure 2). Physical examination was unremarkable and the results of standard laboratory tests were normal. Chest CT scan revealed a tumor that spreads from the lower trachea at the level of the aortic arch to both main bronchi (Figure 3).

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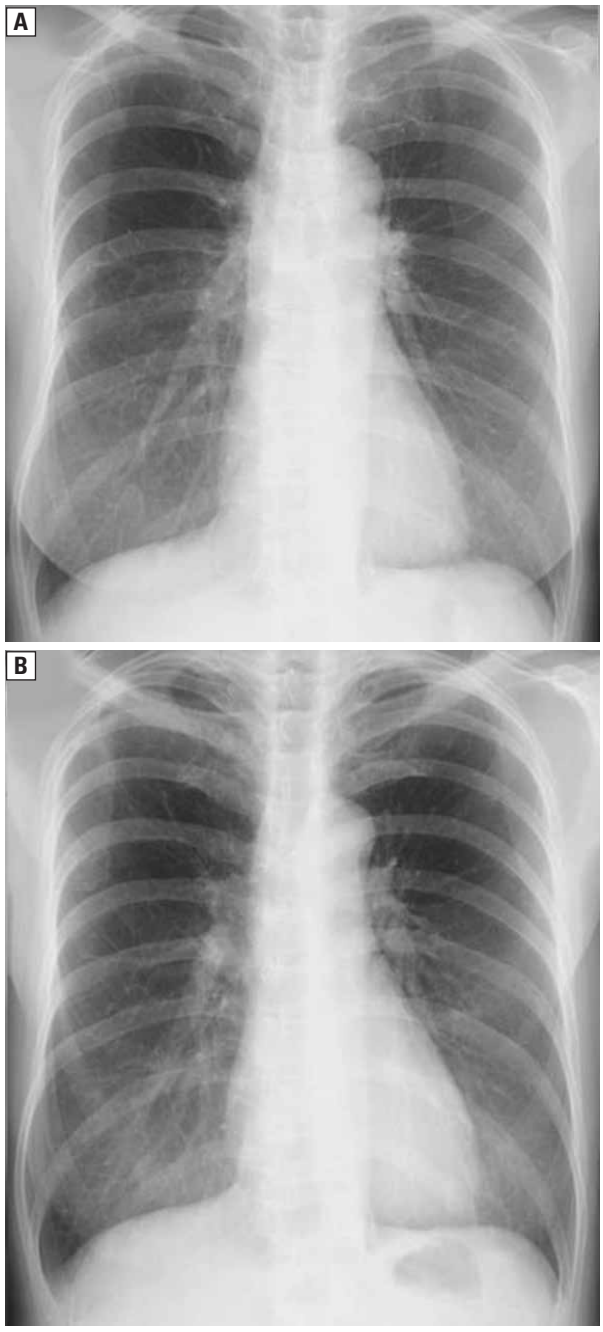


**Figure 1.** In chest radiograph taken this time, the mediastinum structure widened to the right side (**A**) compared to a chest radiograph taken last year for an annual medical check-up (**B**)

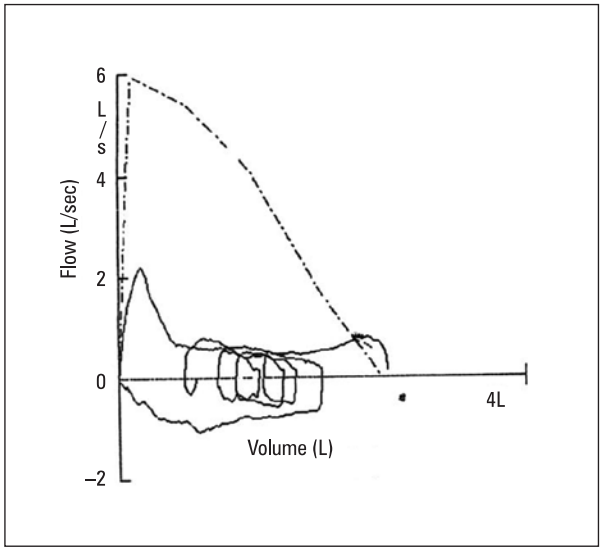


**Figure 2.** The flow–volume curve revealed flat portions of the curve suggesting the central airway obstruction pattern

Bronchoscopy revealed a nodular tumor of almost all circumference with severe stricture of the lumen at the lower trachea (Figure 4). Pathological diagnosis of the specimens obtained from the stenotic lesion was adenoid cystic carcinoma. Proximal and peripheral subepithelial invasion to the middle trachea and the bilateral main bronchi were recognized, therefore, surgical resection was potentially difficult because the extent of resection seemed exceedingly long. An airway stent was inserted, and chemoradiotherapy were performed. The patient is well with no recurrence 4 years after the treatment.

## Discussion

Tracheobronchial tumors are difficult to detect by chest radiograph [4]. This is because these tumors overlap with the structures of the normal mediastinum, vertebral bodies, heart, large blood vessels, and they become unclear. The presence of these tumors is suspected when they extend to the outside of the central airway.

Deformation of the tracheobronchial wall also indicates the presence of tracheobronchial tumors. However, in patients with only thickening of the tracheobronchial wall or those with only extension into the central airway, it is difficult to confirm tracheobronchial tumors by plain chest radiograph. An important point for diagnosis is to notice a few clues and perform chest CT scan.

Workplace-based annual check-up is well established in our country. This annual check-up program has been supported by national government under the Industrial Safety and Health Act. Chest radiograph is an essential in the examination, but the respiratory function test is positioned as optional in this annual check-up program. The pulmonary function test, including the flow-volume curve, is a low-cost test compared to a CT scan and is an easy-to-perform examination in patients with suspected bronchial asthma, chronic obstructive pulmonary disease, and interstitial pneumonia [6–8]. If the presence of these disor-

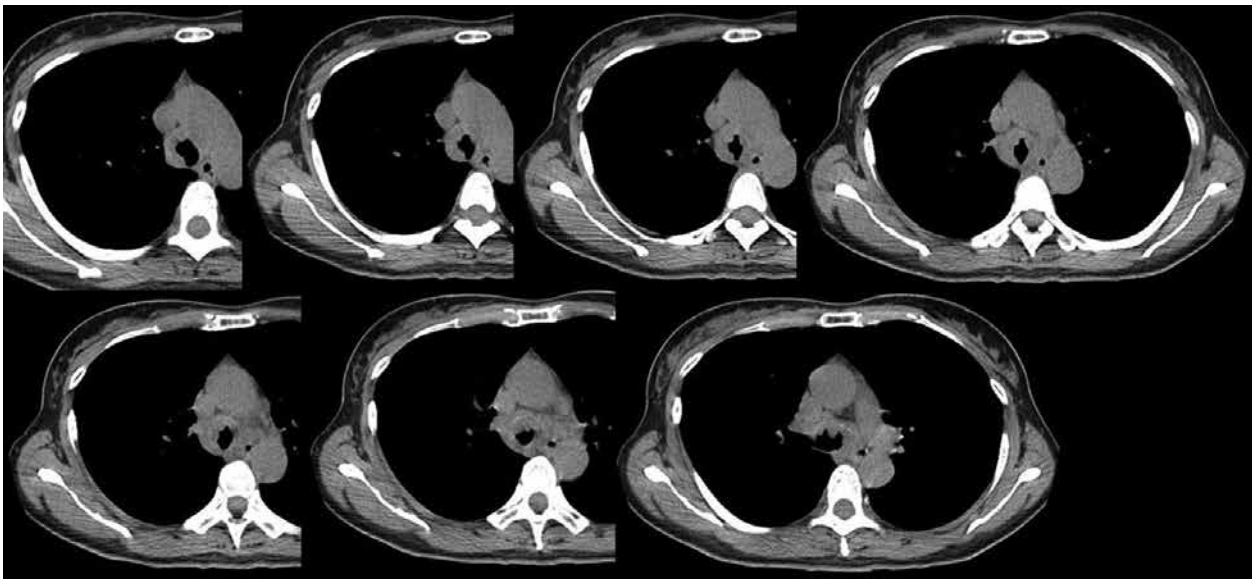

**Figure 3.** Chest computed tomography revealed tumor extended from the lower trachea at level of the aortic arch to both main bronchi

ders is suspected, respiratory function tests can assess the existence of obstructive or restrictive impairment. In addition, the presence of narrowing and stenosis in the central airway can be assessed by the flow-volume curve. The central airway obstruction pattern is characterized by loss of the peak and flat portions of the curve [6–8]. In practice, we might overlook the upper airway obstruction pattern because the peak expiratory flow could be affected by the skill of the laboratory technicians and the effort of the patients. It is therefore important to note whether the procedure was carried out properly.

Primary tracheal tumors comprise about 0.2% of all respiratory tract cancer, with 10–20% and 70–80% being adenoid cystic carcinoma and squamous cell carcinoma, respectively [9, 10]. Adenoid cystic carcinoma originates from secretory glands such as salivary glands and bronchial glands [9, 10]. It accounts for 1% of all cancer cases that originate from the lungs, trachea, and bronchi [9, 10]. There were no reports suggesting a relationship between this cancer and gender and smoking habits [11]. On the other hand, tracheal squamous cell carcinoma is common in men and in patients who have smoking habit [12]. In some patients, the tumors develop along the tracheobronchial wall or into the central airway without extending to surrounding structures. Some patients complain of bloody sputum, and sputum cytology may be useful for diagnosis. For male smokers with bloody sputum, chest physicians perform bronchoscopy. We, chest physicians may be reluctant to perform a bronchoscopy for female

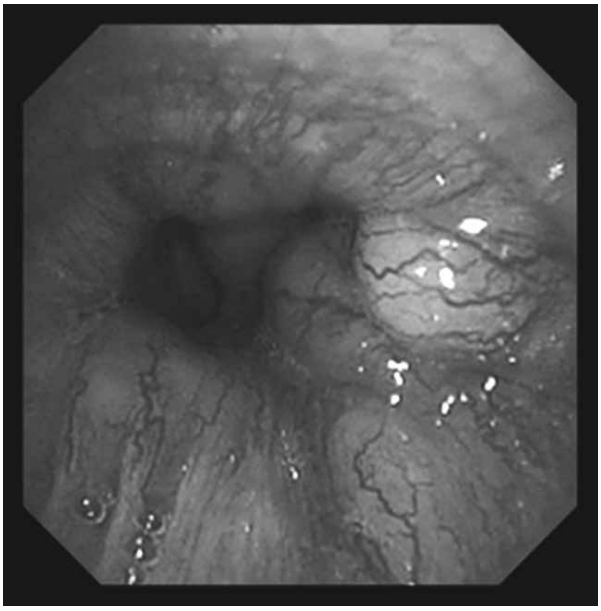

**Figure 4.** Bronchoscopy revealed a tumor of almost all circumference with stricture of the lumen from the lower trachea to both main bronchi. Pathological diagnosis of the specimens obtained from the stenotic lesion was adenoid cystic carcinoma

patients who have never smoked and have no remarkable findings on chest radiograph. Irregularities and deformation of the tracheal wall are typical findings, but may not always be observed in many patients. In our case, the presence of widening of the mediastinum structure to the right side was an important key finding on chest radiograph. Similarly, the flat portion of the curve on the flow-volume loop was an important discovery that should not be overlooked. These subtle obser-

vations led to an accurate diagnosis. A standard treatment for tracheobronchial adenoid cystic carcinoma has not been established. However, early diagnosis of these tumors can be important to initiate most suitable therapies.

Care should be taken not to miss a trivial abnormality in chest radiograph and respiratory function test for early diagnosis. If it is deemed necessary to confirm the findings on these examinations, a chest CT scan should be performed without hesitation.

## Statement of ethics

This study was approved by the institutional ethics committee of each Hospital (Project approval number: NO16-66). Written comprehensive informed consent at the time of admission for obtaining pathological specimens was obtained from the patient.

## Conflict of interest

None declared.

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