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Case report

Tracheobronchopathia osteochondroplastica: Case report and literature review

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A B S T R A C T

Tracheobronchopathia osteochondroplastica (TPO) is a rare disorder characterized as multiple osseous or cartilaginous nodules in the submucosa of trachea and main bronchi. TPO remains an under recognized entity due to lack of awareness. Four cases of TPO are reported in this review as well as various facets of TPO description.

Introduction

Tracheobronchopathia osteochondroplastica (TPO) is an idiopathic non-malignant disease with large airway. Featured by submucosal or osseous nodules overlying the cartilaginous rings, TPO is filled with focal possibility of or danger diffuse. Involved the anterior and lateral walls, nodules sometimes reach main bronchus, even seldom to larynx [1]. In some situation, calcified nodular densities will protrude into the tracheal lumen [2]. However, majority of TPO patients were asymptomatic throughout their lives. The recurrence or chronic resolving pneumonia may appear with the deterioration of airway stenosis accompanied by such symptoms as dyspnoea, hoarseness, persistent, productive cough and haemoptysis [3,4]. Since TPO was first described in details by Wilks in 1867 [5], approximately 400 cases have been reported throughout the world. Here, 4 cases of TPO are presented and make a review of the literature for the various facets of the disease.

Case reports

Case 1

64-year-old male patient was admitted to our clinic research with intermittent fever for 7 months. The patient declared smoking half a packet of cigarettes daily for 20 years. In March, the patient appeared fever with no apparent cause and body temperature reached to 39.5 °C accompanied by chill and cough. The remainder of history was unremarkable. There was no significant abnormality on physical examination. The chest computed tomography (CT) scan showed that the left superior pulmonary was found with inflammatory obstructive. Anti-inflammatory treatments in the early stage were found effective. But such symptoms as fever, cough and expectoration were continued from July to September. Bronchoscopy showed there was distal mucosa hypertrophy in the left main bronchus. One polypoid mass in size of 0.15 cm in the left lingular was observed via opening, while the blade opening was completely blocked. He was admitted to Department of Thoracic Surgery double lingular lobe of left lung cancer. On October 20th, the patient accepted the upper lobe of the left lung resection operation. Histopathologic examination showed that the upper lobe of left lung bronchus with benign lesions, mature bone, cartilage tissue with a nodular hyperplasia sized 1.2*0.9 *0.8 cm. Surface of left lung bronchus was coated with bronchial epithelium, collapse lung tissue ligule, hilar and aortic window reactive hyperplasia of lymph node. So, the situation was diagnosed as TPO.

Case 2

28-year-old male patient was admitted to our hospital with recurrent cough and expectoration for 2 years. Cough and expectoration were aggravated for 1 month before his admission. He smoked 5 cigarettes daily for 3 years. Fever and yellow sputum were found when caught cold. The remainder of history was unremarkable. More physical examination was unremarkable. Anti-inflammatory treatments were effective to relieve symptoms above. Chest CT showed the increasement of tracheal cartilage bone density, the unsmooth lumen and nodular eminence slightly protruded into lumen. Bronchoscopy showed that the tracheal and

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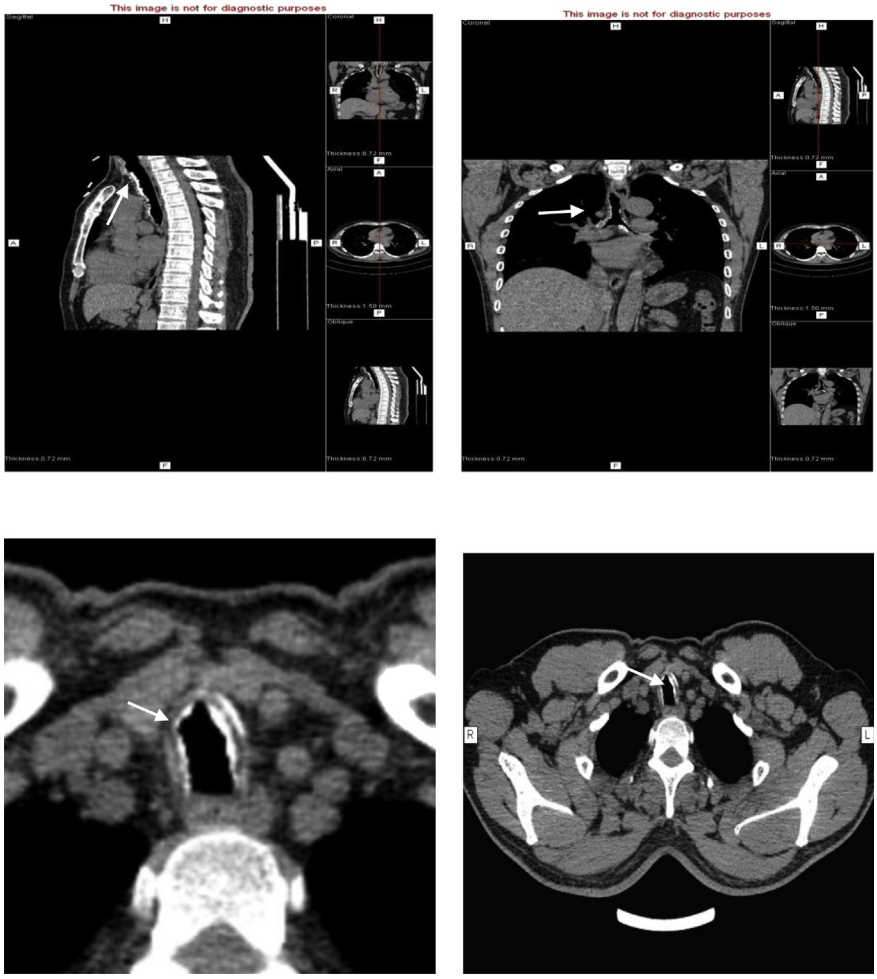

Fig. 1. Chest CT showing the tracheal morphology of the scabbard, irregular thickening and calcified nodules that protruding into the tracheobronchial lumen. Nodules arise from the anterior and lateral aspect of the inner tracheal wall. And the nodules arose from cartilage, posterior membranous wall of trachea is typically spared.

main bronchial cartilages were widely small cobblestone nodular protrusion. Spirometric test was normal. Histopathologic examination of the biopsied tissue showed bronchial epithelial hyperplasia, squamous metaplasia and bone formation. TPO was considered as the cause of chronic cough.

Case 3

45-year-old male patient was admitted to hospital with the productive cough and occasional white sputum for 5 years. The male patient had a past history of pulmonary tuberculosis (21 years ago; unavailable medical records). There was no such history as hemoptysis, loss of appetite, weight loss nor fever. He worked on the highland for long term. The remainder of history was unremarkable. Physical examination was unremarkable. Chest CT scan showed anterior wall of the trachea mucosa was less smooth as well as partial visible flat elevation. Bronchoscopy showed left and front wall surface of tracheal with multiple nodular eminences, hard texture and nodules spread to the carina. Spirometric test showed residual volume to total lung capacity percentage increased slightly. Histopathologic examination showed chronic inflammation of bronchial mucosa with interstitial ossification. The diagnosis was TPO. What is more, during the hospitalization, the patient once appeared sudden abdominal pain, hematochezia. Snap multiple liquid gases flat was found in abdominal plain film. Diagnosis was intestinal obstruction. One colonoscopy check in the mirror at 65 cm showed that a cauliflower like uplift surface hyperemia, erosion pollution moss covered margin irregularly. Histopathologic examination of biopsy sample was reported as a moderately differentiated adenocarcinoma. The patient received surgical operation treatment.

Case 4

49-year-old male patient was admitted with suffered pain in the right side of chest for 2 months. The pain was accompanied by unrelieved right shoulder radiation as well as cough. However, there were no significant incentives when the intermittent pain appeared. One intussusception and partial resection was processed at its 6-month-old. The patient had no history of smoking. The remainder of the history was unremarkable. A slight improvement in symptoms was observed when cefpimizole was given. Physical examination was unremarkable. Chest CT scanning showed scabbard tracheal with calcification, bronchial wall with calcification with irregularly cavity margin (Fig. 1). A subsequent chest CT scan showed no new change. Bronchoscopy revealed widespread mucosal protuberances with white color, which looks like pearl drops in the distal lateral. Anterior walls of trachea were partly conglomerated and swollen mucosal lesions in the right main bronchus, reached to the upper lobe bronchus and similar lesions in

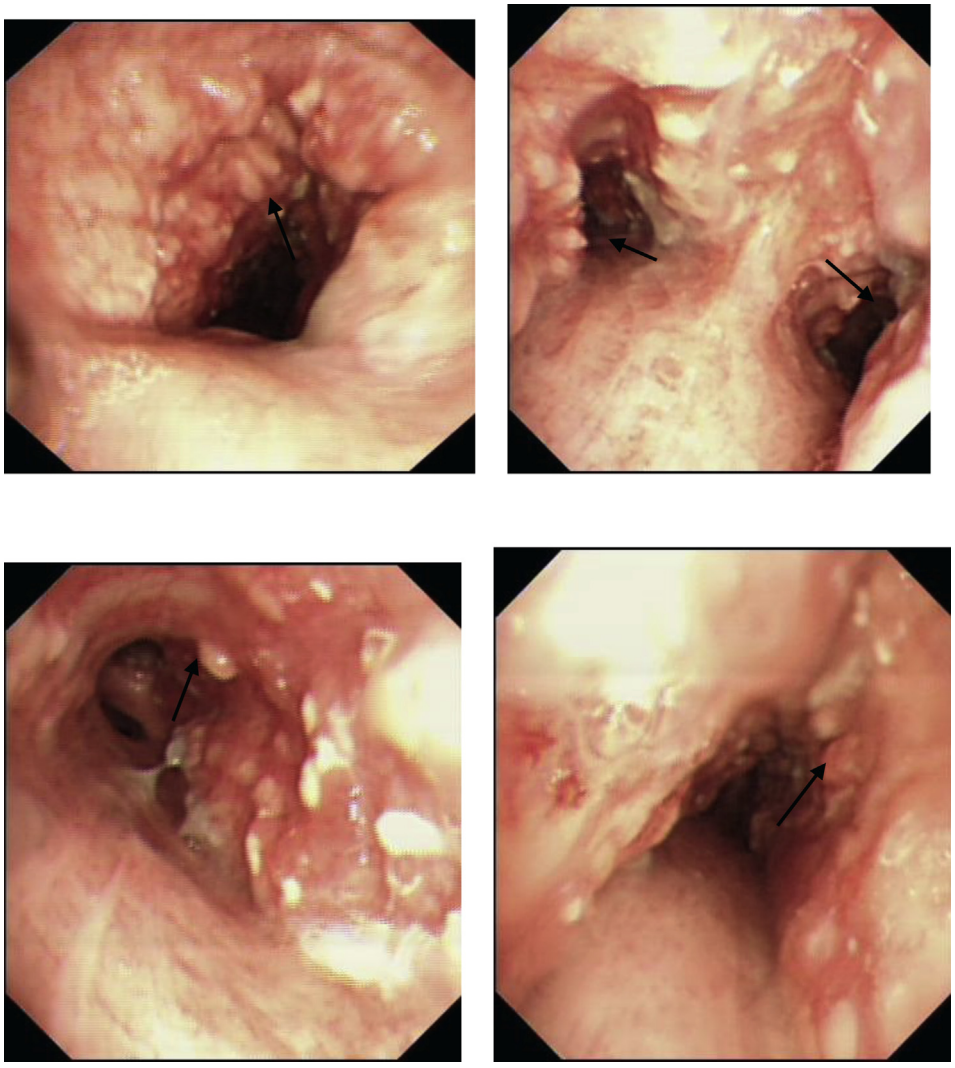

Fig. 2. Bronchoscopy showed widespread mucosal protuberances looking like pearl drops in the distal lateral and anterior walls of trachea. These nodules have "rock garden" or "cobblestone" appearance. Partly conglomerated and swollen mucosal lesions in the right main bronchus reaching to the upper lobe bronchus and similar lesions in the left main bronchus were detected. These nodules are hard on touch and gives gritty sensation while passing the scope through the lumen.

the left main bronchus (Fig. 2). Spirometric test: FEV1 3.12L, FEV1/FVC 72.32L, FVC 4.32L, DLCO SB 11.03 mmol/min/kpa. The arterial blood gas analysis indicated the parameters of hypoxia (PH 7.402, PO_2 79.3 mmHg, PCO_2 38.7 mmHg, HCO_3 23.5 mmol/L, sat 96%). A biopsy sample was taken from the main bronchus. Histopathologic examination showed that the pseudostratified ciliated columnar epithelium mucosa of chronic inflammation with hemorrhage, that epithelium was squamous metaplasia, hyperplasia and

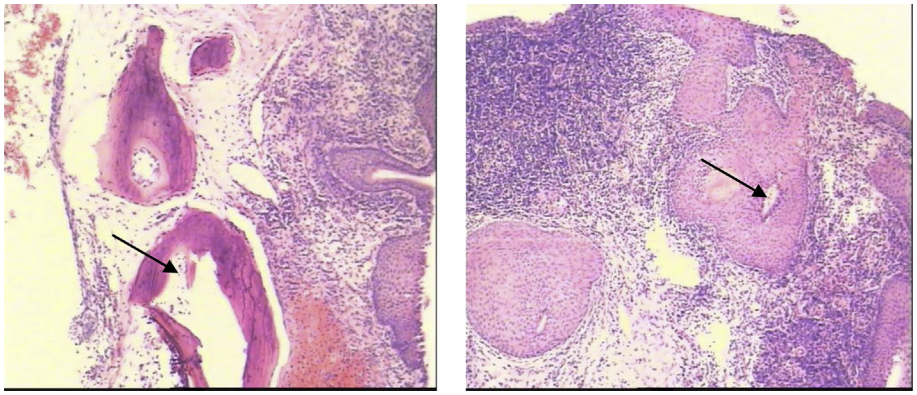

Fig. 3. Histopathological examination showed pseudostratified ciliated columnar epithelium mucosa of chronic inflammation with hemorrhage; epithelium was squamous metaplasia, hyperplasia and hyperkeratosis, submucosa cartilage osseous metaplasia, see lots of keratinized material with calcification.

hyperkeratosis, and lots of keratinized materials with calcification were found within submucosa cartilage osseous metaplasia (Fig. 3). All the features were in line with the characters of TPO.

Discussion

TPO is an uncommon abnormality of the cartilaginous tracheal wall although the actual incidence remains unknown. TPO is more significant serious than we once considered. TPO risk of males is 3 times higher than that of female [6]. It is usually diagnosed in the sixth or seventh decade of life, but one case in a 9-year-old child [7]. In a study of 8760 patients beyond 6 years old who accepted bronchoscopy, 10 cases of TPO were diagnosed, with a median age at diagnosis of 51 years old and male predominance [6]. Chronic infection, congenital anomaly, chemical or mechanical irritation, degenerative or metabolic abnormalities and genetic predisposition are all the possible factors contributing to a person developing TPO [8] although the etiology of TPO is unknown to us.

As we known, TPO is limited to the trachea and main bronchi, but not involve to lung and other organs. The variation of mucosal surface and altered secretions clearance resulted in the recurrence of inflammation and infection. Complaints are varied according to such different accompanied disorder. The major complaints are chronic cough and hemoptysis, even dyspnoea and wheezing occasionally [9].

Diagnosis of TPO is mainly based on computed tomography scanning, bronchoscopy and histopathologic examination. Chest CT scanning showed irregular thickening and calcified nodularties protruding into the tracheobronchial lumen. Nodules were generally arisen from the anterior and lateral aspect of the inner tracheal wall. Although these lesions may extend anywhere from the larynx to the peripheral bronchi, they are more commonly seen at distal 2/3 of trachea and proximal bronchi. Since these nodules arise from cartilage, posterior membranous wall of trachea is typically spared. The sparation is distinguished from such airway diseases as tracheobronchial amyloidosis and Wegener granulomatosis [10]. Bronchoscopy is the most common definitive diagnostic test for these diseases. Bronchoscopy was commonly found as described ‘rock garden’ or ‘cobblestone’ appearance. These nodules were hard to touch and were showed with gritty sensation while the scope passing through the lumen [11]. Further confirmation of the diagnosis is dispensable for either histopathologic examination of the biopsied tissue of the tracheal or bronchial walls at the site of the nodules. The tissue usually showed an osteocartilageous change, with a normal epithelium, whereas cartilage and bone tissue with intervening bone marrow [12]. Amyloidosis, endobronchial sarcoidosis, tuberculosis lesions, papillomatosis, bronchial and tracheal tumors should be considered as alternatively diagnosis for TPO [13].

Treatment is seldom required except in patients with severe airway obstruction and presenting with debilitating symptoms. Conservative therapy aims at maintenance of airway humidity, control of infection and avoidance of airway irritants, treatment modalities include bronchoscopy-guided excision of the nodule, laser ablation, surgical resection and radiotherapy [8,14]. The optimum treatment is still controversial.

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