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A Case of Tracheobronchopathia Osteochondroplastica Associated with Chronic Psoriatic Arthritis in a Patient with Asbestosis Exposure

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Abstract
Tracheobronchopathia osteochondroplastica is characterized by ossified mucosal nodules which protrude into the lumen of the larynx, trachea and bronchi. We report here a 41-year-old man who had psoriatic arthritis for 16 years. He had worked in brake-lining factory for car rim production and brake construction so there he was exposed to occupational asbestos. By fiberoptic bronchoscopy (FOB) there were diffuse mucosal roughening, irregularity and nodularity in the trachea. A biopsy of the subglottic lesions was taken by direct laryngoscopy and diagnosis was made. Although coexistence of this entity with other autoimmune diseases is defined, coexistence with psoriasis has not been previously reported.

Key words: Asbestos, tracheal stenosis, Psoriatic arthritis, Tracheobronchopathia

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INTRODUCTION

Tracheobronchopathia osteochondroplastica or tracheopathia osteoplastica is a rare disorder of the large airways. It is characterized by ossified submucosal nodules which protrude into the lumen of the larynx, and tracheobronchial tree. It has been reported as autopsy findings in a few case reports since 1857, before fiberoptic bronchoscopy (FOB) procedures [1]. The disorder is now encountered more frequently at FOB and/or chest computed tomography (CT) which are usually performed for other reasons [2].

CASE REPORT

A 41-year-old man who had psoriasis for 16 years, was diagnosed as having psoriatic arthritis and indomethacin, and he was administered 15 mg/week methotrexate and methylprednisolon 8 mg/day treatments. At the third month of the treatment he complained of cough, yellow colored sputum, dyspnea with exertion, and a foreign body sensation in his throat. In the physical examination, he had psoriatic skin lesions on his back and especially on knees, elbows, hands and feet. He had no clubbing. The remainder of his exam was normal. He had no other disease in his previous history but he had 8 pack-year history of cigarette smoking which he quitted 16 years ago. He had worked in brake-lining factory for car rim production and brake construction so there he was exposed to occupational asbestos. Spirometry showed no abnormality with a forced expiratory volume in one second (FEV_1) of 2.25 L(80%) and forced vital capacity (FVC) of 2.90 L(89%); the ratio of FEV_1/FVC was 78%. Pulmonary diffusing capacity (DLCO/VA) was 5.82 mL/mmHg/min/L (94%). Routine laboratory test results were within normal limits except for a erythrocyte sedimentation rate of 59 mm/h, and the WBC count of 13100/mm3. Rheumatoid factor was 34 U/L and antinuclear antibody (ANA) was positive with nucleolar painting.

The chest radiograph revealed interstitial reticular changes of both lungs. A chest high resolution computed tomography (HRCT) scan showed an increase in anteroposterior diameter of trachea, narrowing of lumen and a nodularity in the mucosa protruding through the lumen (Figure 1). The horizontal diameter which was narrowed at the upper part of the trachea was seen by spiral three dimensional CT. By fiberoptic bronchoscopy (FOB) there were diffuse mucosal roughening, irregularity and nodularity in the trachea just distal of the vocal cords which continued up to middle 1/3rd of trachea (Figure 2). Vocal cord mobility was normal. Because of severe narrowing in the airway it was not possible to pass distally with FOB. Histopathological analysis of biopsy taken from tracheal lesions revealed that mucosa was lined with stratified squamous epithelium and submucosa involved with an active granulation tissue rich in vascular structures.

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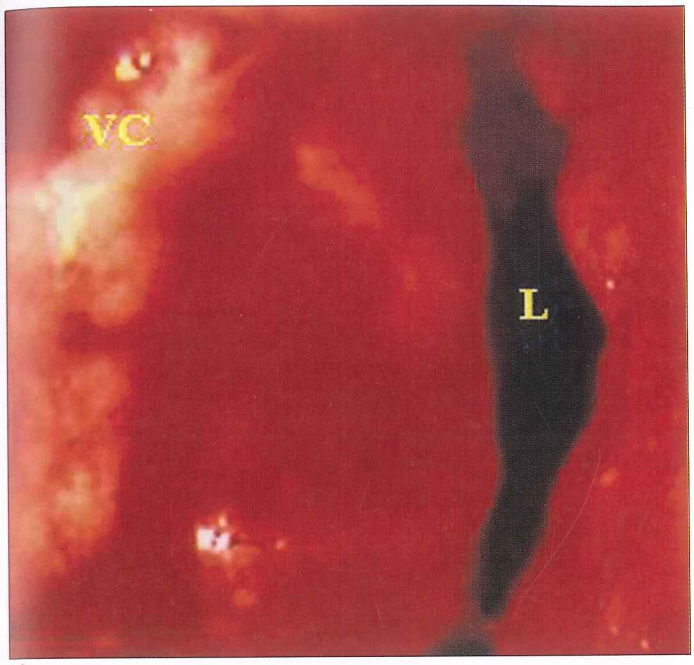

Figure 1. CT scan shows the mucosal irregularity with calcification in anterior and lateral walls of the trachea. The posterior membranous wall is intact
L: lumen of the trachea, VC: vocal cord

By spiral CT scanning, the contour irregularity, and surrounding calcification at the trachea and two main bronchi were observed. Although the patency of the lumen was distally seen, the horizontal diameter was narrowed at the upper part of the trachea (Figure 2). In the cervical CT which was taken because of dysphonia, pathological cervical and supraclavicular lymphadenopathy was not present. In the area from the ventricular level to thoracic aperture for trachea, there were some nodular calcifications in the tracheal wall.

In direct laryngoscopy, a polyp was observed on anterior side of the vocal cords explaining the cause of dysphonia. During the intubation with a size 8.0 cuffed tracheal tube, there was difficulty in progression of the intubation tube. Tracheal intubation was performed with a smaller size endotracheal tube (inner diameter, 7.0 mm). A biopsy of the subglottic lesions was taken with some difficulty because of their bony nature.

In histopathological examination, the tissues consisted of multiple fragments of mucosal tissue lined by respiratory or metaplastic squamous epithelium. Within the lamina propria, calcified foci of bone tissue were noted (Figure 3). No amyloid were observed in tissue sections stained by Congo-red. The clinicopathological diagnosis was that of Tracheobronchopathia osteoplastica.

DISCUSSION

Tracheobronchopathia osteoplastica (TO) is a rare disease of unknown cause affecting the laryngotracheobronchial tree. It is characterized by cartilaginous or bony outgrowths into the lumen of the tracheobronchial tree as seen by bronchoscopy. Although the true incidence of TO is not known with certainty, the frequency of appearance in routine bronchoscopic examinations was reported to be one in 2000-5000 bronchoscopies [3]. Appearance of TO was found higher at autopsies as it is generally asymptomatic. In one report, TO was an incidental finding at autopsy in 1 in 400 cases [4].

It may be clinically asymptomatic as it is in our case. And sometimes it is an incidental finding that is noticed during difficult intubation in surgical procedures [5-7]. Symptomatic TO is accompanied by cough, hoarseness, hemoptysis, exertion dyspnea and wheezing. The cough is thought to result from turbulent air flow in the trachea, increased airway sensitivity, and impaired ciliary clearance caused by nodules. Physical examination is unremarkable in most cases. Stridor and ronchi may occur when severe airway obstruction is present, and a clinical picture simulating asthma has been described [2].

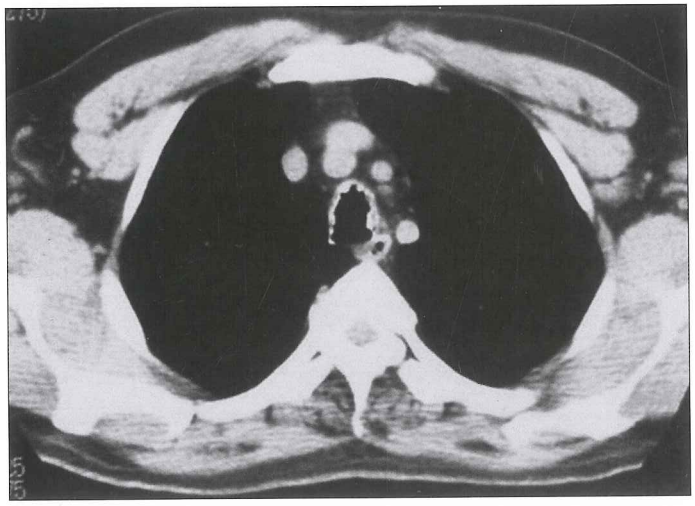

Figure 2. Mucosal roughening, irregularity and nodularity was seen in the trachea just distal of the vocal cords by fiberoptic bronchoscopy

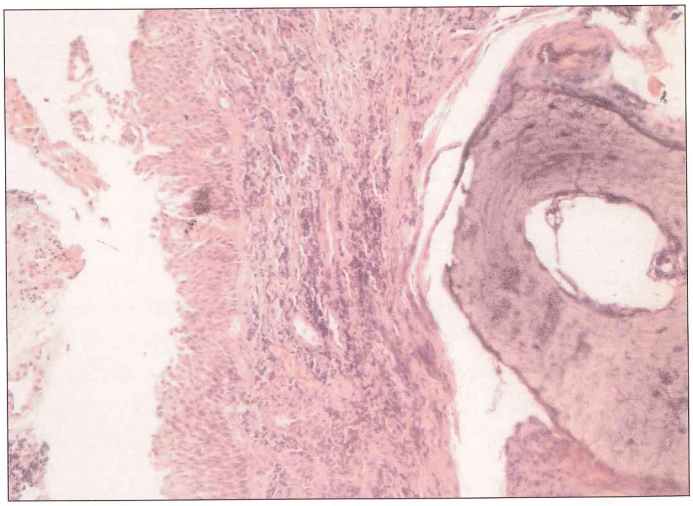

Figure 3. Calcified bone tissue within the lamina propria. H&E, original magnification x20

Chest computed tomography demonstrated dense submucosal nodules mainly in the trachea and main bronchi. They are calcified in half of cases as in our case. Usually the lesions appear at distal two-thirds of the trachea and major bronchi but they may be found at proximal part of trachea and larynx as in our case. Although involvement of the larynx is reported as a rare finding, Nienhuis et al. demonstrated upper airway involvement including larynx and upper trachea abnormalities in 40% of their patients [8]. Krenke et al. notified a small soft nodule in the larynx besides the typical nodules protruding into the lumen of trachea and main bronchi [9]. Histological examination showed a polyp with regions of inflammation and necrosis. We have also seen a laryngeal polyp in the present case by direct laryngoscopy.

It is diagnosed when nodules with "cobble stone" appearance are seen protruding into the lumen of trachea and large bronchi [2,10]. However, this similar appearance can also be detected in papillomatosis, amyloidosis, calcifying lesions of tuberculosis and endobronchial sarcoidosis [1,5,11]. Definitive diagnosis may be made by bronchoscopy, computed tomography or magnetic resonance imaging. While there is no thickening of tracheal wall in the above mentioned entities where calcification occurs in tracheal cartilage, there is no cobble stone appearance in posterior mucous membrane in TO which indicates that membrane is preserved.

Etiology is not clear but chronic disease, chemical and mechanical irritation, degenerative or metabolic abnormalities and genetic predisposition are being accused [5]. The coexistence of TO and atrophic rhinitis has been described in several instances. Although the bacteria Klebsiella ozenae is frequently isolated in both conditions, which suggests some link between these disorders [2,5], there is no clear relationship demonstrated so far [2].

Although coexistence of this entity with other autoimmune diseases such as dermatomyositis and scleroderma is defined, coexistence with psoriasis has not been previously reported. Additionally, our case has occupational asbestos exposure and TO may be developed in chronic psoriasis base due to chronic irritant effect to asbestos.

The mechanisms of nodule formation in TO are unknown. Various hypotheses have been postulated: According to Virchow, the nodules represent exostoses and ecchondrosis from the cartilaginous trachea which subsequently undergo calcification and ossification. Aschoff-Freiburg postulated that the disease is the result of abnormalities in the tracheal elastic tissue. Furthermore, Dalgaard stated that undifferentiated connective tissue cells develop into cartilage cells through true metaplasia in the internal layer of the elastic connective tissue of the surface layer of the submucosa and lamina propria. These cells lose their immature character and calcium accumulates in them and the intercellular space to form bone tissue [2,5,10].

No objective data support these hypotheses. Recent immunohistochemical studies of TO lesions have suggested a role for bone morphogenetic protein 2 (BMP-2), a member of the transforming growth factor-b family which plays important physiological roles in the formation of new bone and cartilage [10,12]. There is no known genetic susceptibility for the development of TO.

The disease usually remains stable for years, or progresses very slowly. Only a minority of cases develop significant upper airway obstruction and require invasive procedures to remove or bypass the obstruction on affected airways. To date there is no specific treatment for the disease. In the patients experiencing frequent infections and/or severe airway obstruction, cryotherapy, excision with Nd:YAG laser or external radiation may be carried out or the obstructive lesion may be removed on bronchoscopy. Although the disease known to be benign, deaths due to bronchial obstruction have been reported [13].

In conclusion, the occurrence of TO in the present case may be the result of the metaplasia caused by a chronic irritant effect of occupational asbestos exposure, on the basis of an autoimmune disease.

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