Tracheobronchopathia Osteochondroplastica in a Patient with Silicosis

CT, Bronchoscopy, and Pathology Findings

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Abstract: A case of tracheobronchopathia osteochondroplastica in a patient with silicosis is reported, showing a rare association of disease. Etiological hypotheses and clinical aspects are discussed. Radiologic, bronchoscopic, and pathologic findings are demonstrated with emphasis on the role of computed tomography (CT) in the diagnosis of this disease.

Key Words: tracheobronchopathia osteochondroplastica, silicosis, imaging, bronchoscopy

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Tracheobronchopathia osteochondroplastica (T.O.) is a rare disease characterized by multiple cartilaginous or bony nodules of various sizes involving the lateral and anterior walls of the trachea.[1] The nodules are composed by bone or calcified acellular protein matrix.[2] The disease occurs more commonly in males over 50 years,[3] and its prevalence ranges from 0.02 to 0.7% in routine bronchoscopies.[4] The bronchoscopic findings consist of multiple irregular protrusions, hard in texture, whitegriseous in color, which have described as resembling a rock garden by Gautam in 1968.[2–5] CT plays an important role in the recognition of this entity. The characteristic CT findings consist of calcified nodular densities protruding into the tracheal lumen and resulting in an irregular shape of the tracheal contour and decreased lateral diameter.[6] We report the clinical, CT, bronchoscopic, and pathologic findings in one case of T.O. in a patient with silicosis.

CASE REPORT

A 50-year-old man presented with productive cough, blood-tinged sputum, and dyspnea on exertion for 2 years. Physical examination disclosed diminished breath sounds and sparse rhonchi. The patient had worked in a foundry and was exposed to silica dust for 22 years. There was no history of tuberculosis or other infectious diseases in the past.

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Chest radiographs revealed discrete calcification and narrowing of the trachea, bilateral nodular infiltrates with apical coalescence compatible with complicated silicosis (small opacities "qr," profusion 3/2, large opacities "C," according to ILO classification)[7] (Fig. 1).

A CT scan was performed, and showed thickened tracheal cartilage with calcification of the nodules leading to narrowing of the tracheal lumen (Fig. 2). CT also demonstrated characteristic parenchymal findings of silicosis with conglomerate upper lobe masses of fibrosis, bilaterally.

The patient underwent a fiberoptic bronchoscopy, which revealed numerous hard and pale nodules along the anterolateral wall of the trachea (Fig. 3). The diagnosis was confirmed by examination of biopsy specimens, which showed connective tissue lined by ciliated columnar cells and areas of calcification in the submucosa (Fig. 4). Amyloid stain was negative. There was no evidence of autoimmune disease. Sputum, bronchoalveolar lavage, and a biopsy did not present evidence of malignant tumor cells. Cultures for *Mycobacterium tuberculosis*, bacteria, and fungi were negative.

The patient died a few months after the diagnosis due to a severe pneumonia.

DISCUSSION

The first description of T.O. was by Walks, who in 1857 observed ossific deposits protruding into the lumen of larynx, trachea, and bronchi in an old man who died of tuberculosis.[3] Although the disease is uncommon, it is probably underdiagnosed.[8]

Many hypotheses have been postulated regarding the etiology of this disease, such as infectious, postinfectious, posttraumatic, inflammatory, idiopathic, and inhalation of toxic substances. Nevertheless, its etiology is still unknown.[9] Only 2 case reports describe T.O. associated with pneumoconioses.[10,11] Jepsen and Sorensen reported a case of T.O. and chronic atopic rhinitis in a patient who was exposed to oil vapors and suggested that chronic irritation was responsible for both conditions.[2] The occupational exposure to silica for many years could have acted as a chronic irritant, perhaps contributing to the development of the tracheal lesions.

This disease is usually benign, and the patient can be asymptomatic and frequently is diagnosed incidentally during intubation or bronchoscopy.[12] The most common symptoms are cough, sinusitis, hemoptysis, hoarseness, dyspnea, and wheezing.[13] Respiratory infections and acute dyspnea by extension to the larynx or significant narrowing of the tracheal lumen can occur in the course of the disease.[14]

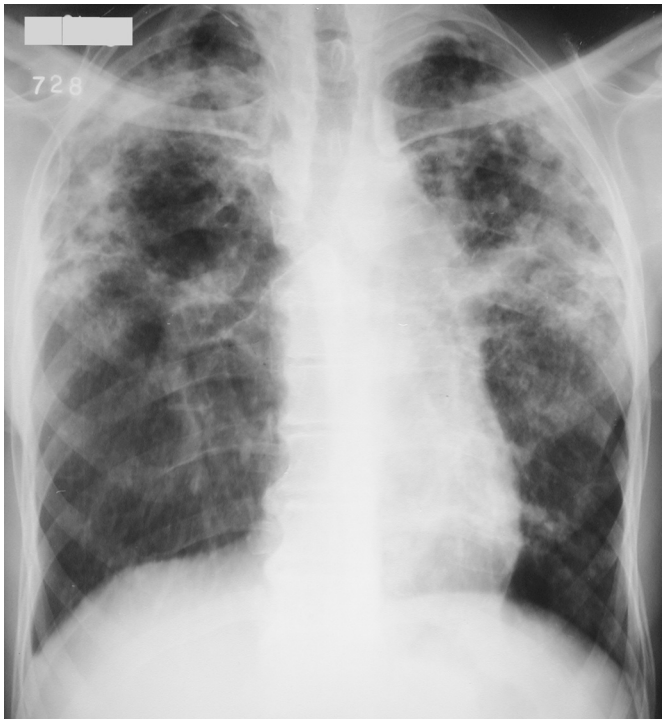

FIGURE 1. Chest radiograph shows numerous upper lobe nodules consistent with silicosis, superior retraction of the hila, and upper lobe bullae. Also, note slight irregularity of the tracheal lumen.

The differential diagnosis includes amyloidosis, cancer, endobronchial sarcoidosis, calcified tuberculosis, papillomatosis, and Wegener granulomatosis.[3,15,16]

In our study, CT correlated very well with bronchoscopy, and histopathology confirmed the diagnosis. The CT findings

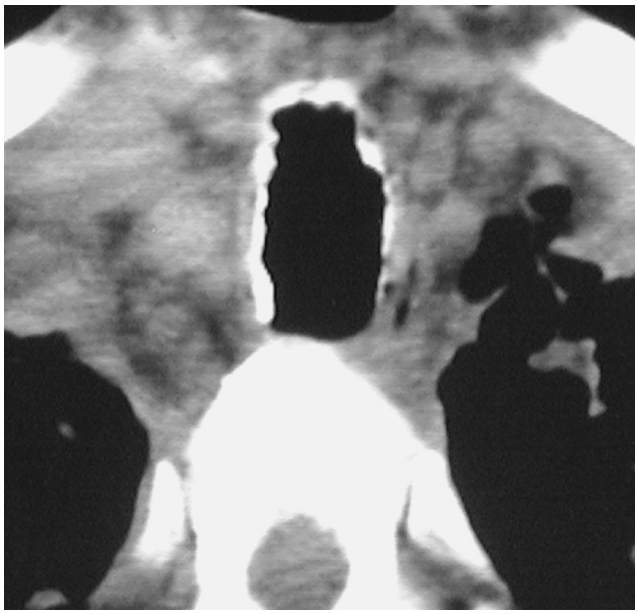

FIGURE 2. CT scan (10-mm collimation) shows extensive nodular calcification of the anterior and lateral tracheal wall.

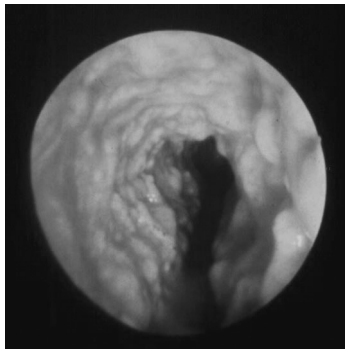

FIGURE 3. Bronchoscopic image revealing numerous nodules on anterolateral wall of the trachea.

can be considered highly sensitive for the diagnosis of this disease, with the advantage that it is a noninvasive method, which improves the detection and characterization of central airway disease.[4] Magnetic resonance imaging (MRI) can demonstrate the irregular thickening of the trachea and its extent, but it is less sensitive than CT for demonstrating the punctiform calcifications in the submucosal nodules and the potential complications, such as lobar collapse and bronchiectasis.[17] The introduction of new image technologies including multidetector spiral CT, which allows endoscopic 3D processing of image in the investigation of T.O., will probably enhance the diagnosis in the future.[1,18] Nevertheless, bronchoscopy remains the primary procedure for the diagnosis of T.O.[4]

The prognosis of T.O. is usually favorable and no treatment is required, unless for symptom relief, or infections, when antibiotics may be used. In severe cases of the disease, the removal of nodules with laser or endoscopy surgery can be attempted but, according to Prakash et al, this approach did not produce satisfactory results. Surgery may be indicated in some cases. Successful segmental tracheal resection and temporary insertions of a stent have been reported.[15,17,19]

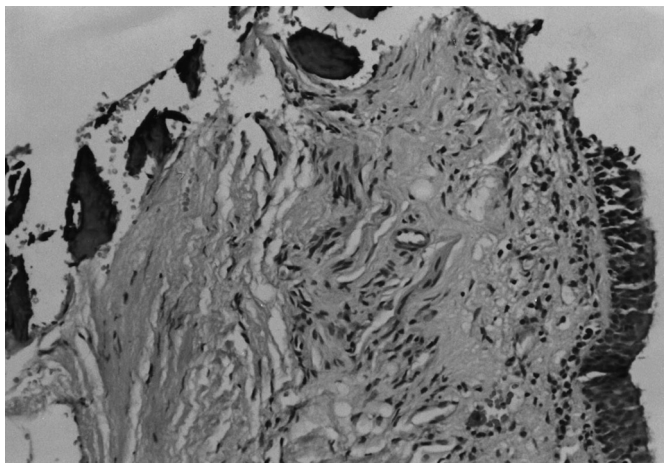

FIGURE 4. Bronchofiberscopic biopsy of trachea showing areas of ossification in submucosa (hematoxylin and eosin, ×40).

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