Atypical Presentation of Tracheobronchopathia Osteochondroplastica: Is Chronic Inflammation a Perpetrator?

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Abstract

**Objective:** To report an atypical presentation of tracheobronchopathia osteochondroplastica (TO). **Clinical Presentation and Intervention:** A 59-year-old man was investigated for productive cough of 1 month. An antimycobacterial combination regime was initiated with a misdiagnosis of endobronchial tuberculosis. At follow-up, the patient reported worsening of his symptoms. CT revealed an increased intensity of the cartilage ring surrounding the trachea, and bronchoscopy showed tracheal stenosis with white, hard nodules on the airway submucosa. Histopathology confirmed the diagnosis of TO. **Conclusion:** This case showed that TO should be considered in patients with cough not explained by non-invasive testing and not responsive to empiric medications.

Introduction

Tracheobronchopathia osteochondroplastica (TO) is characterized by development of multiple osseous and cartilaginous nodules in the submucosa of the trachea and the main bronchus [1, 2]. Patients usually present with cough, recurrent respiratory tract infections, and occasionally hemoptysis [3, 4]. TO is not usually suspected until fiber-optic bronchoscopy is performed; the bronchoscopy views together with histopathological examination of the nodules confirm the diagnosis.

Case Report

A 59-year-old male smoker (30 pack-years) was investigated for productive cough of 1 month. He had a past history of pulmonary tuberculosis (25 years ago; unavailable medical records) and mentioned occasional episodes of dry cough every year for the last 3 years. His family and occupational history were not significant. One month earlier, he had been diagnosed with pulmonary tuberculosis at another institution, but his sputum smear and culture...
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Discussion

TO is limited to the large airways and does not involve the lung or other organs [1]. Changes at the mucosal surface and altered clearance of secretions result in recurrent inflammation and infection [3]. These lesions typically spread over the anterior and lateral walls of the airways (but not the posterior wall). Studies suggest that only 51% of patients with TO are accurately diagnosed during their lifetime [5]. Our patient was reevaluated after 1 month, CT and bronchoscopy revealed features suggestive of TO, but acute deterioration is not typical of this disease [6]. Classic hypotheses include ecchondrosis and exostosis arising from the cartilaginous rings, or metaplasia of the submucosal elastic and connective tissue [1–3]. An association with lung cancer (adenocarcinoma in particular) has also been suggested [7]. Cystic fibrosis coupled with bacterial infection induces metaplastic bone replacement, as well as destruction and elimination of the bronchial cartilage. Degenerative changes in the cartilage and increased perichondrial fibrosis have been demonstrated in patients with chronic obstructive pulmonary disease and bronchial asthma. We assume that chronic inflammation of the large airway, in part due to recurrent infection, may have been the cause for our patient’s initial complaint of cough. Moreover, the acute changes in the airways observed 1 month after starting the antitumor combination regime and the symptomatic recovery after discontinuation of the drugs are an interesting association that remains unexplained. Whether the antitumor drugs played a synergistic role by accelerating the inflammatory process is debatable. There are reports suggesting coexistence of tracheobronchial amyloidosis and TO [8]. Isolation of Klebsiella pneumoniae, both in atrophic rhinitis and TO, suggests a link between these disorders [9]. Other differential diagnoses of TO include calcified lesions secondary to tuberculosis, carcinoma, papilloma, fibroma, endobronchial sarcoïdosis, polychondritis, and Wegener’s granulomatosis of the proximal airways. Immunohistochemical studies of TO lesions suggest a role for bone morphogenetic protein 2 [10]. The above-mentioned features were not evident in our patient, and the short history made understanding his case rather complex. Multiple factors are probably involved in the pathogenesis of TO; the cartilage ossification seen in our pa-
tient may possibly be the result of an intense inflammatory reaction in the bronchial mucosa. More case reports and studies on the etiology of the condition will help to clarify this issue.

There is lack of consensus among clinicians on the optimum treatment, while 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 [1–4].

**Conclusion**

This case report showed that TO should be considered in patients with cough not explained by noninvasive testing and not responsive to empiric medications. CT results may be suggestive, but bronchoscopy examination, followed by histopathological findings is diagnostic of TO. Interventional bronchoscopy has an important role in the symptomatic treatment of TO.

**References**