CASE REPORT

Tracheobronchopathia Osteochondroplastica and Lung Carcinoma: A Rare Combination

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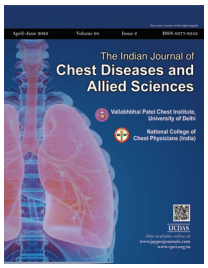

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Abstract

Tracheobronchopathia osteochondroplastica is a rare benign idiopathic disorder of unknown cause affecting the large airways. Bronchoscopy and computed tomography (CT) of the thorax establish the diagnosis, but biopsy is confirmatory. We describe a case of tracheobronchopathia osteochondroplastica, rarely associated with lung cancer. There should be a high index of suspicion of tracheobronchopathia osteochondroplastica, especially in patients with malignancy; as it can easily be missed in a patient with lung malignancy.

Keywords: Bronchoscopy, Lung neoplasm, Osteochondrodysplasias.

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Abbreviations used in this Article

CT = Computed tomography; TTF-1 = Thyroid transcription factor 1

Introduction

Tracheobronchopathia osteochondroplastica is a rare idiopathic disease of the tracheobronchial tree. Most often, it is an incidental diagnosis and warrants conservative management as its course is self-limiting. In symptomatic individuals, it resembles airway diseases, like chronic obstructive pulmonary disease, asthma, and sarcoidosis.[1] The etiology of tracheobronchopathia osteochondroplastica remains unknown; chronic inflammatory process, degenerative pathology, metabolic, or neoplastic pathology are postulated.[2] Computed tomography and flexible bronchoscopy appearance of tracheobronchopathia osteochondroplastica are pathognomic of the diagnosis. However, confirmation of the diagnosis with histopathological sampling may be required.

Case Description

A 69-year-old woman with a suspected left upper lobe lung mass presented to the outpatient department with a dry cough. She was a nonsmoker and had no history of chronic respiratory diseases or any other comorbidities. General physical examination showed no clinical abnormalities. There was early clubbing and no associated lymphadenopathy. Past medical history was unremarkable. Computed tomography of the chest showed diffuse, irregular nodules, and calcification of the tracheal wall along with left upper lobe mass (Fig. 1). Bronchoscopy revealed multiple, hard irregular nodules protruding into the tracheal lumen sparing the posterior wall of the trachea without any obvious endobronchial lesions (Fig. 2). In view of the age, a high suspicion of lung cancer was considered as these lesions were postulated to be malignant deposits. Multiple transbronchial lung biopsies were taken from lung mass and tracheal lesions. Histopathological analysis of lung mass was suggestive of adenocarcinoma of the lung. Immunohistochemistry was positive for thyroid transcription factor 1 (TTF-1) and positive for epidermal growth factor receptor mutation analysis. Biopsy from tracheal lesions showed tissue lined by respiratory epithelium, subepithelium having osteocartilaginous tissue with chronic inflammatory cells, and fibrosis compatible with tracheobronchopathia osteochondroplastica without any malignant cells (Fig. 3)

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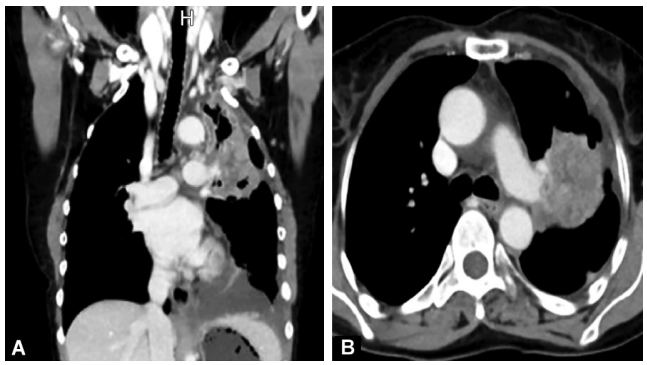

Figs 1A and B: Chest computed tomography (coronal reconstruction) showing thickening and calcified lesions of the trachea and left upper lobe cavitating mass lesion

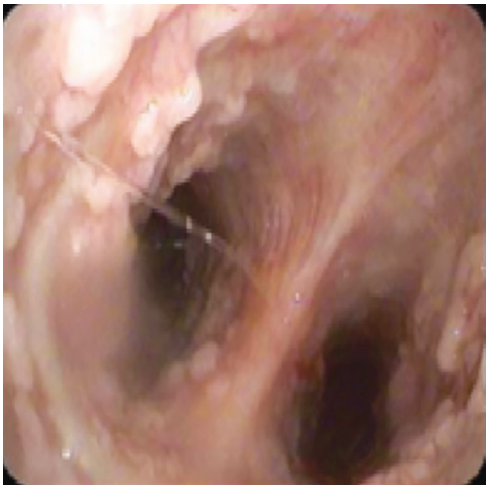

Fig. 2: Bronchoscopic view showed multiple, irregular nodules in the trachea and the main bronchi, sparing the posterior wall

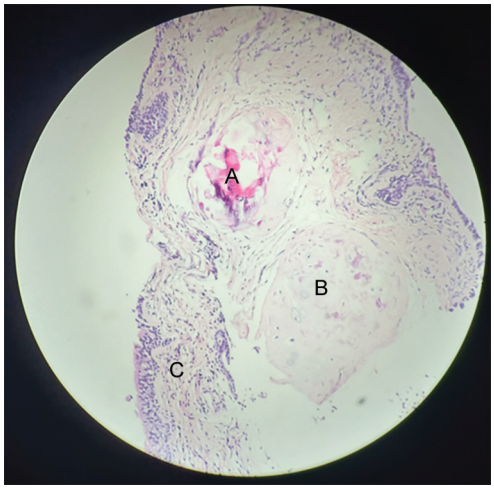

Fig. 3: Histopathological examination showed tissue lined by pseudostratified columnar epithelium. (A) Subepithelium showed bone, (B) Cartilaginous tissue, and (C) Fibrosis and chronic inflammatory cells

Discussion

Tracheobronchopathia osteochondroplastica is one of the rare disorders of benign etiology characterized by multiple cartilaginous and osseous nodules in the sub-mucosal region projecting into the lumen of the respiratory tract. Tracheobronchopathia osteochondroplastica usually occurs in individuals of more than 50 years, but it is also been described in children. There is no sex predilection and no clear association with smoking and other occupational diseases.[3] Tracheobronchopathia osteochondroplastica is asymptomatic but can present with wheezing, cough, hemoptysis, dyspnea, and rarely obstructive symptoms. Wilks described this condition in 1857, who found sub-mucosal projections covered by bony plates in the larynx, trachea, and bronchi during autopsy studies.[4] Chest radiograph is usually normal. Danckers et al.[5] first reported this condition with a mass lesion, which was causing acute dyspnea secondary to obstructive atelectasis. In a few cases, it can be associated with lobar collapse and post-obstructive bronchiectasis. Detection and diagnosis of tracheobronchopathia osteochondroplastica by a CT scan requires careful interpretation of the findings and biopsy for confirmation. Tracheobronchopathia osteochondroplastica was an incidental diagnosis, but an increasing number of cases are being diagnosed in recent years because of advanced diagnostic procedures, such as bronchoscopy or CT. Few cases of tracheobronchopathia osteochondroplastica are diagnosed after difficult intubation.[6] Currently, most cases are diagnosed during bronchoscopy and CT. Tracheobronchopathia osteochondroplastica appear as whitish, hard spicules projecting into the tracheal lumen from the anterior and lateral walls during bronchoscopy and autopsy, sparing the posterior wall. The larynx and the main bronchi also can be involved in a few cases. In our case, the chest radiography revealed a left upper lobe cavitating mass, but the CT showed multiple nodular densities in the trachea apart from cavitating mass in the left upper lobe, which led to the suspicion of tracheal metastasis. Bronchoscopy from lung mass and the speculations of the trachea showed findings that were characteristic of tracheobronchopathia osteochondroplastica. Histological examination of the nodules revealed varying combinations of cartilaginous, osseous, and hemopoietic tissue within a calcified protein matrix, protruding into the bronchial lumen consistent with tracheobronchopathia osteochondroplastica. The overlying mucosa is often the site of squamous metaplasia.[7] Lesions of the tracheobronchopathia osteochondroplastica are not pre-malignant. This progress slowly into the tracheal lumen and rarely need treatment. In chronic cases of tracheobronchopathia osteochondroplastica, obstruction and recurrent infections might occur which requires urgent intervention. There is no definitive therapy and most of the cases need only symptomatic treatment, which includes antibiotics in case of secondary bacterial infections. Very few cases require mechanical measures to debulk nodules using external beam irradiation, radiotherapy, cryotherapy, laser excision, stent insertion, or surgical resection therapy.[8–10] The differential diagnosis to the bronchoscopic picture of multiple nodules includes tracheobronchopathia osteochondroplastica, amyloidosis, endobronchial sarcoidosis, calcific tuberculosis, papillomatosis, and tracheobronchial calcinosis.[11] The etiology of tracheobronchopathia osteochondroplastica remains unclear. Some hypotheses include exostoses acquired metaplasia, ossification of cartilage, and reactive inflammation secondary to chronic infection. The occurrence of the disease in some familial cases suggests that there has also been a genetic predisposition that needs to be proven. A few cases have been associated with lung cancer and a few with thyroid tumors or thymoma.[12] Some reports suggested that lung cancer was found in 11.1% of patients associated with tracheobronchopathia osteochondroplastica. Tracheobronchopathia osteochondroplastica has been associated with several pulmonary neoplasms; most notably adenocarcinoma, mucoepidermoid carcinoma, non-Hodgkin's lymphoma, and there is no clear genetic correlation between tracheobronchopathia osteochondroplastica and lung cancer.[13] Association with malignancy can be just coincidental.[14] Reasons for this co-existence may be because of common risk factors like smoking.[15] In our patient, tracheobronchopathia osteochondroplastica was associated with adenocarcinoma lung. In conclusion, tracheobronchopathia osteochondroplastica in association with lung cancer has been reported rarely and histological confirmation is needed for the diagnosis. If tracheobronchopathia osteochondroplastica is not suspected as a differential diagnosis of malignant invasion of the trachea, it can easily be misdiagnosed as metastasis, especially in patients associated with the malignancy.

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