Case Report

Tracheobronchopathia Osteochondroplastica: A Rare Cause of Difficult Intubation

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Abstract

Tracheobronchopathia osteochondroplastica is a rare benign airway disorder which is characterised by submucosal nodules projecting into the tracheo-bronchial lumen usually involving the cartilaginous portions of the tracheo-bronchial tree or larynx. The condition is usually asymptomatic but can rarely present with difficulty during endotracheal intubation or rarely with obstructive airway complications. Bronchoscopic appearance is usually sufficient to make the diagnosis, and tissue biopsies are seldom required. No specific treatment is required in asymptomatic patients. However, interventional bronchoscopy procedures or surgery may be helpful in symptomatic cases. **[Indian J Chest Dis Allied Sci 2014;56:187-189]**

Key words: Tracheobronchopathia osteochondroplastica, Ossification, Airway obstruction.

Introduction

Tracheobronchopathia osteochondroplastica (TPO) is an unusual and rare benign airway disease, which is characterised by bony or cartilaginous projections into the lumen of larynx, trachea or bronchi, with sparing of the posterior membranous trachea. Computed tomography (CT) may reveal beaded calcification of the tracheo-bronchial cartilages. On histopathological examination, the abnormal growths show heterotopic bone formation. Diagnosis is usually delayed because of the chronic and asymptomatic nature of the condition. Symptoms may include dyspnoea, chronic cough, haemoptysis, hoarseness of voice, and wheezing. Diagnosis is made on the basis of bronchoscopy findings, CT and histopathology. No known treatment is available for this condition. Surgical treatment is indicated for airway obstruction and recurrent infections.[1]

The condition may be diagnosed incidentally during tracheal intubation or bronchoscopy. We report one such instance when difficult tracheal intubation was encountered in a patient who was undergoing surgery for rectal carcinoma. The diagnosis of TPO was suspected on CT of the thorax and was subsequently confirmed with flexible fiberoptic bronchoscopy and histopathological examination.

Case Report

A 40-year-old man was evaluated in the surgical out-patient clinic for complaints of altered bowel habits and was diagnosed to have carcinoma of the rectum. Patient had no history of dyspnoea, haemoptysis, chronic cough or fever. His past medical history and family history was unremarkable. He was a life-time non-smoker with no known occupational exposure. Pre-operative spirometry and chest radiograph were normal. Abdomino-perineal resection for rectal carcinoma was performed. During tracheal intubation for general anaesthesia, the patient could not be intubated with 7.5mm size endotracheal tube. With difficulty, intubation was performed with 6.5mm sized endotracheal tube. At the time, anaesthetist noticed some nodules just below the vocal cords. The surgery was completed successfully and the post-operative period was also uneventful.

After post-operative recovery, the patient underwent evaluation for the cause of difficult intubation and an opinion from the pulmonary medicine services was sought. Post-operatively, examination of the respiratory and cardiovascular systems was normal. Computed tomography of the thorax (Figure 1) demonstrated calcific nodular and beaded appearance of the anterior and lateral tracheal walls with calcified nodules protruding into the tracheal lumen. Flexible fibreoptic bronchoscopy (Figure 2) showed extensive variable sized nodules on the anterior and lateral walls of trachea and both proximal mainstem bronchi which were protruding into the airway lumen and causing narrowing of the proximal tracheo-bronchial tree. Sparing of the posterior membranous wall of the trachea was observed. Nodules had stony hard feel when closure was attempted after grasping with biopsy forceps.

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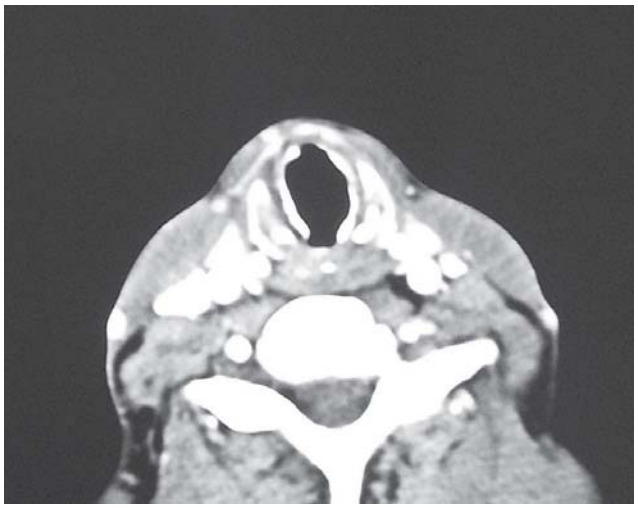

Figure 1. Computed tomography of chest demonstrating nodular calcified appearance of the anterior and lateral tracheal walls leading to a "beaded" appearance.

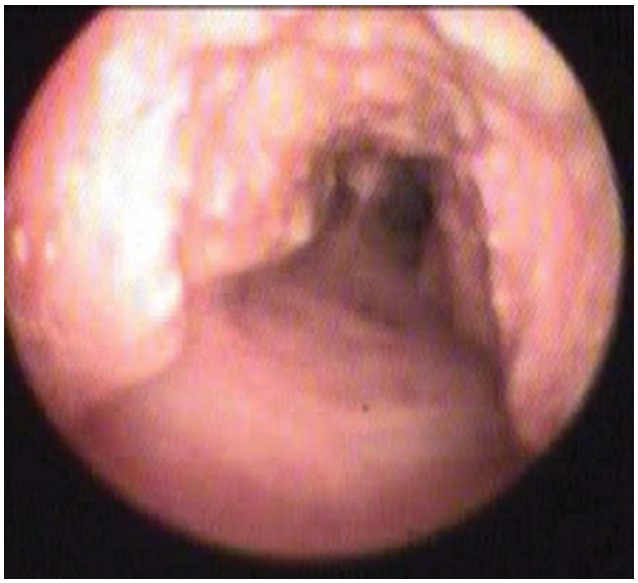

Figure 2. Flexible bronchoscopy demonstrating extensive nodularity of the entire trachea in its entire extent and carina with sparing of the posterior membranous trachea.

Histopathological examination of the nodule biopsy specimen (Figure 3) showed respiratory lining epithelium with underlying sub-epithelium showing dense calcification. There was no evidence of malignancy, granulomatous inflammation or amyloid deposition. In view of the classical CT and flexible bronchoscopy examination findings and the histopathology examination results, diagnosis of TPO was confirmed. Our patient was asymptomatic, spirometry was within normal limits [forced expiratory volume in the first second (FEV_1) 2.61 Litres (81.3% predicted); forced vital capacity 3.42 Litres (84.2% predicted); and FEV_1/FVC 76.5%] and shape of the flow volume loop was normal (Figure 4).

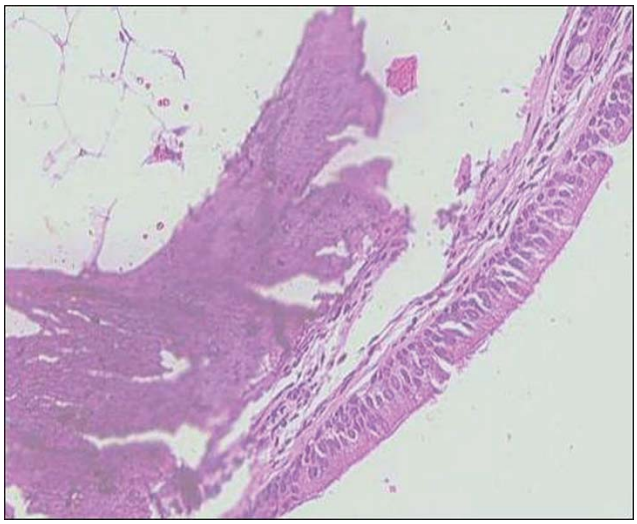

Figure 3. Photomicrgraph of histopathological examination from one of the tracheal nodules demonstrating dense sub-epithelial calcification confirming the diagnosis of tracheobronchopathia osteochondroplastica (Hematoxylin and Eosin × 100).

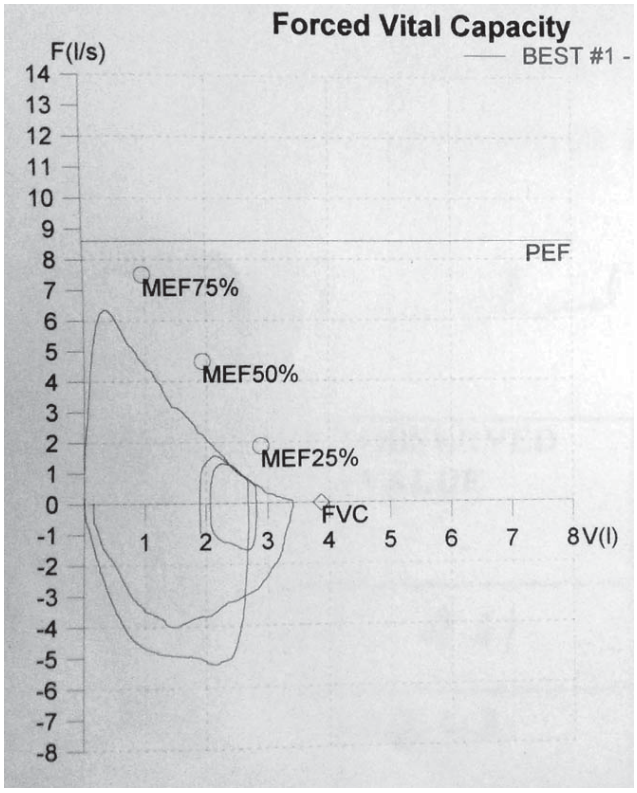

Figure 4. Photograph of the flow-volume loop obtained during spirometry examination.

Follow-up with periodic clinical examination and spirometry was planned. Patient was counselled to voluntarily disclose his underlying condition, should he ever undergo any surgical procedure in the future requiring administration of general anaesthesia.

Discussion

The underlying aetiology and pathogenesis of TPO is unknown. Chronic infections (e.g., mycobacterial), chemical exposure or mechanical irritation (e.g., silicosis), metabolic abnormalities (e.g., amyloidosis) and genetic predisposition are some of the proposed causal factors.[2-4] Congenital predisposition has also been suggested. Reported incidence is approximately 3/1000 at autopsy while that reported in bronchoscopic data range from 1/125 to 1/5000 procedures.[5] Most commonly, TPO is diagnosed in patients aged over 50 years. Although no gender predominance has been described, some reports suggest that males are affected more commonly.[6] Clinically, majority of the patients remain asymptomatic. However, the nodules in TPO may be large enough to significantly impinge on the tracheo-bronchial lumen and the presentation is related to the degree of the airway obstruction and the level of obstruction (glottis, sub-glottis or the tracheo-bronchial level). This condition is usually discovered incidentally during bronchoscopy or at autopsy or during difficult intubation as in the index patient.[7,8] Prior to the advent of flexible bronchoscopy, a large proportion of the cases were diagnosed at post-mortem examination.[5]

In our patient, an epiglottic nodule was also observed. However, there was no significant glottic or tracheo-bronchial obstruction. No characteristic pattern of pulmonary function test pattern has been described in this condition. Lung function tests may be normal as was seen in our patient.[9] Flow-volume loops may be a more sensitive indicator of disease progression. With severe airflow obstruction, the flow-volume loop is characterised by a plateau/flattening in both the inspiratory and expiratory limbs of the curve as this condition physiologically leads to a fixed airway obstruction. Chest radiograph is usually normal as in our patient. The CT findings suggestive of TPO include thickening and irregularity of the tracheal and/or bronchial walls with nodular calcified protrusions into the lumen as were evident in our patient. Flexible fibreoptic bronchoscopic examination is the most useful tool for establishing the diagnosis. The characteristic bronchoscopic findings as observed in the index patient include whitish, hard spicules that impart a cobble-stoned appearance to the airway wall (Figure 2). Tissue sampling is seldom required to confirm the diagnosis of TPO and obtaining a bronchoscopic forceps biopsy may prove to be difficult owing to the stony hard nature of the nodules. In situations with atypical clinical or bronchoscopy features, tissue biopsy must be obtained. Histopathological examination from the bronchoscopic biopsy specimens when available, displays heterotopic bone formation with abnormal cartilage proliferation and calcium deposits.

Differential diagnoses include other luminal narrowing conditions, like tracheo-bronchial amyloidosis, endobronchial sarcoidosis, calcifying endobronchial tuberculosis, tracheo-bronchial granulomatosis with polyangitis, relapsing polychondritis and primary neoplasms of the airways.

Generally, TPO is considered as a benign airway disorder and asymptomatic patients could be simply followed for prolonged periods of time. When the trachea or the bronchi are significantly narrowed, surgical treatment (tracheal or laryngeal resection) and bronchoscopic therapy (removal of nodules with biopsy forceps, laser ablation and cryotherapy) have been attempted. External beam radiation has also been tried with reported relief of symptoms.[10]

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